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Transcatheter embolization of abnormal intrathoracic vessels using coils in the setting of children with congenital heart disease

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Abstract

Objective: it was the purpose of this retrospective study to assess the efficacy and the rate of complications of transcatheter embolization of abnormal intrathoracic vessels using coils in children with complex congenital heart disease.

Patients and methods: in 17 children (mean age 9.2±5.9 years) with complex congenital heart disease, occlusion by catheter intervention was attempted in 29 abnormal intrathoracic vessels: 13 aorto-pulmonary collaterals, 12 arterio-pulmonary collaterals, 2 systemic arteries supplying pulmonary sequestrations, one central venous connection and a Blalock-Taussig shunt. The mean diameter of the vessels was 4.7±1.6 mm (range 2-8 mm). Steel coils with a helical diameter of three, five or eight mm were used. After selective catheterization of the vessel, they were delivered through a 5 F endhole catheter. The helical diameter of the coils was chosen in order to exceed the inner diameter of the vessel by 10-30%.

Results: as assessed by selective angiography performed 10 minutes or more after release of the coil, 27 of the 29 vessels (93%) were successfully occluded. A mean of 2.6 coils (range 1-11) were necessary for successful occlusion. Complications were encountered during 4 attempts of occlusion (14%). Of a total of 76 coils delivered, 4 coils (5%) secondarily migrated after release from the catheter, mostly to branches of the pulmonary arteries. This complication was seen predominantly in those vessels with the largest diameters. Three of the dislocated coils were left in place as they did not significantly obstruct flow of blood. One coil was retrieved by a basket catheter.

Conclusion: transcatheter embolization of abnormal intrathoracic vessels using coils in children with congenital heart disease is an effective therapy. Potential complications warrant careful evaluation of the indications for these procedures.

Key words: Aortopulmonary collaterals, transcatheter embolization, children

Since the first description in 1975 of occlusion of vessels by mechanical devices introduced through catheters,[1] several authors have shown the feasibility and effectiveness of embolization with steel coils for a wide variety of intrathoracic vessels and vascular malformations in children with congenital heart disease.[2,3] The complication rate was low throughout all the reports. Experience with this intervention in children, however, is still limited. This retrospective study was undertaken to assess the role of the characteristics of the vessels on the rate of success and on the frequency of complications when embolization was performed in children with complex congenital heart disease.

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Methods

Between February 1987 and October 1994, transcatheter embolization was attempted for 29 abnormal intrathoracic vessels in 17 children with

congenital heart disease. Embolizations were performed during 22 catheterizations (1-3/child). Of the catheterizations, 17 were routine pre- or postoperative hemodynamic evaluations. In these cases, the indication for occlusion of the vessel was based on the immediate results of the hemodynamic calculations and on angiography. The other 5 catheterizations were performed with the sole purpose of embolization of already known abnormal vessels. All intrathoracic systemic-to-pulmonary collateral arteries with significant flow and size (greater than 2-3 mm in diameter) were considered an indication for occlusion.

Preparation and monitoring of the patients did not differ from routine catheterization. The premedication was usually oral diazepam. Besides local anesthesia of the groin (prilocaine), the older children received sedation with morphine (0.05-0.1 mg/kg per dose) whereas younger children received ketamine (1-2 mg/kg per dose). A standard percutaneous femoral arterial and venous approach was used in all patients. Intravenous heparin (100 U/kg) was given after arterial access had been obtained. Before delivery of the coils, and for 24 hours thereafter, all patients received an intravenous antibiotic (cefazoline).

Seven vessels in six patients (five aortopulmonary collaterals, one Blalock-Taussig-shunt and one central venous connection) were occluded as part of the preoperative management before corrective or palliative cardiac surgery. These occlusions were performed after discussion with the surgeon. The intention was to prevent bleeding complications and to occlude vessels that would have not been reached easily by the surgeon during the operation. In a patient with Scimitar syndrome, two systemic vessels supplying pulmonary sequestrations were occluded as the first therapy, preceeding surgical closure of the patent ductus. In 10 patients, 20 embolizations of systemic-to-pulmonary collateral arteries were performed as part of the postoperative management (Table 1).

The detailed anatomy and geometry of the vessels to be occluded were determined by selective angiography. The inner diameter of the vessels was measured on the angiographic films at different sites. Presence of distal stenosis was of special interest. The site of embolization was chosen depending on the course of the vessel, but was usually in the proximal part of the vessel shortly after branching off the feeding systemic artery.

MWCE 38 steel coils (Cook, Europe) were used in all instances. These coils consist of surgical stainless steel supporting thrombogenic Dacron strands. The extended diameter of the coils was 0.97 mm (0.038 inch), and the extended length was 3, 4 and 5 cm, respectively. The diameter of the coiled emboli was 3, 5 or 8 mm, respectively. The first coil used was chosen so that the coiled diameter of the embolus was about 10-30% larger than the inner diameter of the vessel as measured on angiographic film. The MWCE 38 coils were delivered with the use of 5F endhole catheters. The type of catheter chosen varied according to the site and anatomy of the target vessel. Catheters were used that best allowed selective catheterization of the ideal site of occlusion. The extended coils were introduced into the catheter once selective catheterization had been achieved. Coils were pushed forward and released from the catheter with a 0.97 mm (0.038 inch) flexible guide wire.

Table 1. Embolization procedures in relation to cardiac diagnosis of patients

Diagnosis	Number of patients	Vessels embolized preop.	Vessels embolized postop.	Failure of embolization
Complete transposition:				
s/p ASO	1		1	
s/p Mustard	3		4	
s/p Rastelli	1		5	
Scimitar-syndrome	1	2		
DILV/Fontan	2	1	4	
Ebstein	2	2		
DILV	1	1		1
TOF/PAtr	5	3	5	1
PAtr/IVS	1		1	
Total	17	9	20	2

ASO = arterial switch operation, DILV = double inlet left ventricle, IVS = intact ventricular septum, PAtr = pulmonary atresia, s/p = status postoperative, TOF = tetralogy of Fallot

Occlusion was expected to occur by thrombosis within 10 minutes of embolization. Success of the intervention was assessed by angiography not earlier than 10 minutes after delivery of the coils. Total occlusion was defined as lack of flow of contrast past the site of embolization. Subtotal occlusion was defined as minimal residual flow past the coils, with only faint opacification of the vessel distal to the site of embolization. Unsuccessful embolization was defined as clear visualisation of the distal course of the vessel by contrast medium. In such instances, further coils were delivered to achieve at least subtotal occlusion.

As quantification of shunting is impossible in patients with multiple aortopulmonary collateral

arteries, complete hemodynamic assessment after embolization was not routinely repeated, and only arterial saturations were obtained. All children routinely had a chest X-ray the day after the intervention for exclusion of pulmonary infiltrations and pleural effusions. Recatheterization in the follow-up after embolization was performed only if indicated for hemodynamic evaluation of the underlying heart disease or prior to reoperation, but in that case the result of embolization was assessed by angiography.

Statistical values were expressed as mean ± standard deviation. Differences between mean values were compared using the unpaired t-test. A p-value of <0.05 was considered statistically significant.

Results

The age of the patients varied from 0.2 - 18.3 years (mean 9.2 ± 5.9 years). Only one patient was aged less than one year at the time of the procedure. In this patient with Scimitar syndrome two systemic vessels supplying large pulmonary sequestrations were occluded. Two more patients were in the second year of life. Occlusion of a large indirect aorto-pulmonary collateral artery (originating from the right subclavian artery) one year after an arterial switch operation for transposition was performed in the first of these patients and in the second patient with tetralogy and pulmonary atresia a large aorto-pulmonary collateral artery was occluded preoperatively. Two patients, who were more than 16 years old at the time of occlusion were included in the study because they were still cared for by the pediatric cardiologists due to the complexity of their malformations.

Weight at intervention varied from 4 - 82 kg (mean 34.3 ± 11 kg). Ten children were male, seven female. All the patients had complex congenital cardiac malformations (Table 1).

In 11 patients, one vessel was occluded, while in 3 children two vessels were treated, and in 3 children three, four and five vessels were embolized, respectively. Of the vessels, 13 were direct aortopulmonary collateral arteries and 12 were indirect aortopulmonary collateral arteries originating from one of the major aortic branches, mainly subclavian arteries. Two vessels were systemic arteries supplying sequestered segments of lung and, in one instance, a Blalock-Taussig-shunt and a central venous connection (azygos vein to pulmonary venous confluence) were embolized, respectively. The diameter at the origin of the vessels varied from 2 to 8 mm (mean 4.7 ± 1.6 mm). Distal narrowing was present in 15 of the vessels where occlusion was attempted but 14 vessels showed approximately the same diameter throughout their course, as seen on angiography,without significant narrowing in the distal parts of the vessel.

Of the 29 embolizations, 27 (93%) were successful. Immediate evaluation by angiography more than 10 minutes after embolization showed total occlusion (Fig.1) in 13 vessels (45%), and subtotal occlusion with only minimal residual flow in 14 (48%). In 6 of the vessels with only subtotal occlusion, angiography during recatheterization of these patients after 1 - 34 months (median 11 months) post-embolization then showed total occlusion of all 6 vessels. In no case was dilation found proximal to the site of occlusion during these follow-up studies.

The mean diameter of the vessels that showed total obstruction to flow after embolization was 3.9 ± 1.1 mm (range 3 - 6), whereas the mean diameter of the vessels with minimal residual flow at post-interventional angiography was 5.1 + 1.3 mm with a range of 2 - 7 mm (p=0,04). The mean diameter of those three vessels where dislocation of coils occurred was 6.7 ± 1.5 mm (range 5 - 8 mm) (p=ns). All the 14 vessels with a diameter equal to or less than 4 mm could successfully be occluded, whereas embolization failed due to coil migration in 2 of 15 vessels with a diameter greater than 4 mm (Table 2). In those vessels where total occlusion was achieved, a mean of 3 coils (1 - 7) had been placed. In the vessels with subtotal occlusion, a mean of 2.5 coils had been placed (p=ns).

The presence or absence of stenosis of the distal part of the vessel did not seem to influence the rate of success (one failure or complication in 15 embolization attempts in vessels with stenosis as compared to two failures or complications in 14 attempts in vessels without stenosis) (Table 2). In the 27 successful embolizations, between 1 and 11 coils (mean 2.6 ± 2.3) had been delivered (76 in total). There was no correlation between diameter and the number of coils necessary to achieve total or subtotal occlusion of blood flow.

The main complication observed was migration of the coil after its release from the catheter into the vessel. This was seen in four attempts (three vessels). The diameter was greater than 5 mm in all three children. In a 15-year-old boy with double inlet left ventricle and discordant ventriculo-arterial connections, an attempt was made to occlude a Blalock-Taussig-shunt prior to reoperation. Two consecutive attempts of embolization resulted in dislocation of the 8 mm

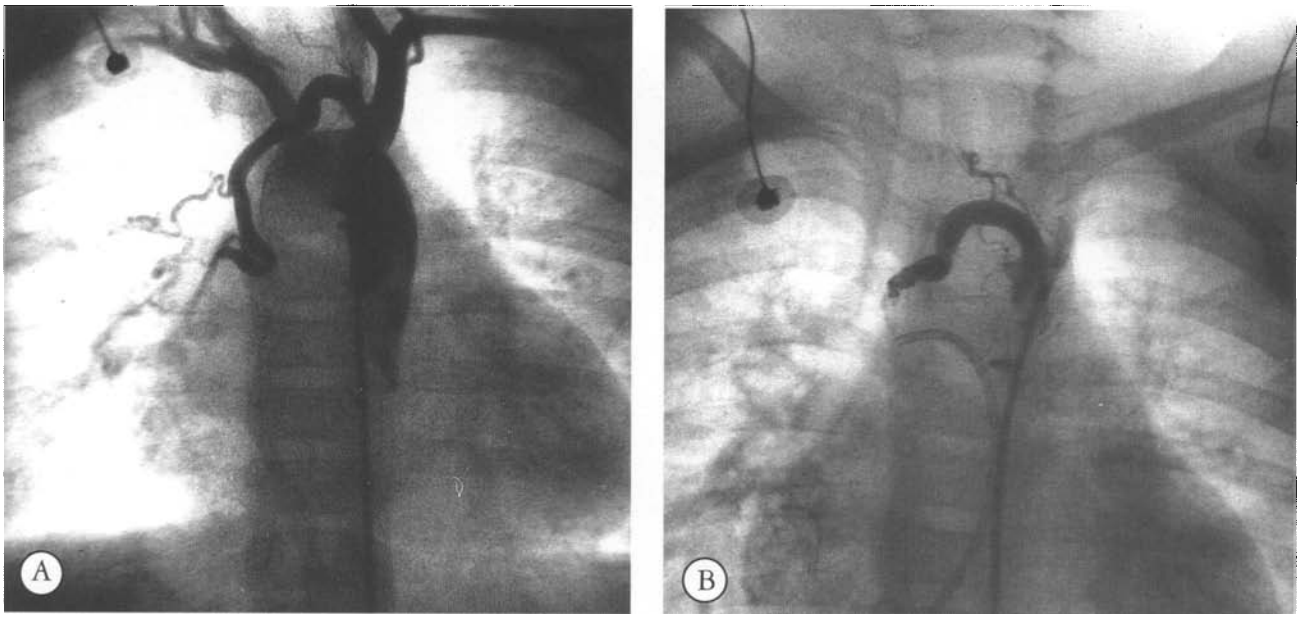

Figure 1.
A). Systemic-to-pulmonary collateral vessel (4 mm in diameter) in a 15 month old boy after neonatal arterial switch operation for simple transposition. B). After delivery of 3 coils no residual flow is seen in the angiography 15 minutes after embolization (total occlusion).

Table 2. Relation between success rate of embolization and vessel characteristics

Result	Distal vessel stenosis		Vessel diameter	
	present	absent	≤ 4mm	4>mm
Successful embolization	14/15	13/14	14/14	13/15
Coil migration	1	2	0	3*

*dislocation of 4 coils in 3 vessels

coil to a branch of the right pulmonary artery. The devices were left in place, as they did not significantly obstruct blood flow as evaluated by pulmonary angiography. Recatheterization after 6 months confirmed patency of that pulmonary artery. An 11-year-old girl who had a Fontan-type operation, had developed collateral arteries from the left subclavian artery to the left pulmonary artery. One coil had already been placed successfully when a second 5 mm coil was delivered. This coil migrated to the subclavian artery and was successfully caught with a retrieval basket, but had to be removed from the right iliac artery by arteriotomy. In a 5-year-old girl with tetralogy and pulmonary atresia, an 8 mm coil migrated from a major aortopulmonary vessel (7 mm diameter) to a branch of the right pulmonary artery after release from the catheter. Angiography showed no significant obstruction to flow and the coil was left in place. Pulmonary angiography 7 years later confirmed patency of that vessel (Fig.2). In a 5-year-old boy who had had a Mustard operation for complete transposition, a coil could not be released from the catheter. At retrieval of the whole system, the coil finally separated from the catheter in the right iliac artery and had to be removed with a retrieval basket. The collateral vessel was subsequently successfully embolized during the same session.

In seven children, pulmonary perfusion was assessed (by angiography in six patients and by scintigraphy in one child) 7 months to 3 years (mean 17 months) after successful embolization of a systemic-to-pulmonary collateral. In no instance was there a localized perfusion deficit near the site of embolization.

Except for one 17-year-old patient, who experienced transient chest pain after occlusion of a collateral vessel from the right internal thoracic artery, no child complained of side effects from the procedure. No child developed pleural effusions or signs of pulmonary infarction, as assessed by clinical examination and chest X-ray the day after the intervention and before hospital discharge.

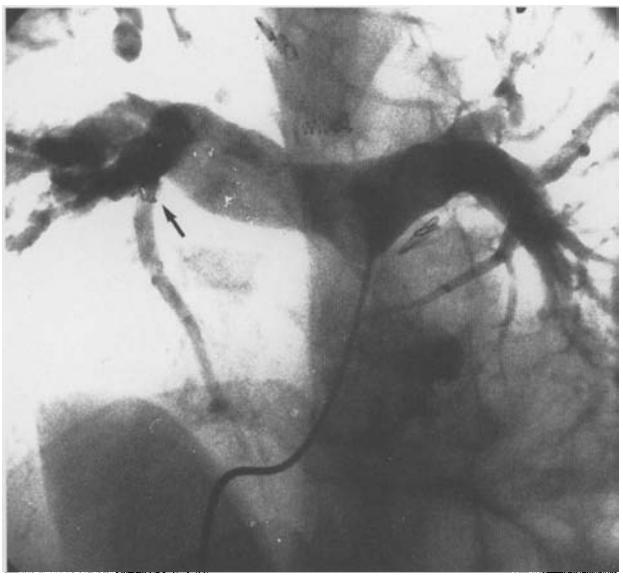

Figure 2.
12-year-old girl with pulmonary atresia and ventricular septal defect. Multiple coils are seen in the region of the aortic arch after prior successful occlusion of major aortopulmonary collaterals. Migration of a coil to a right branch pulmonary artery occurred at an attempt of embolization of another collateral. The coil was left in place (arrow). The figure shows the result of pulmonary angiography 7 years after coil dislocation occurred: no significant obstruction to flow in that branch pulmonary artery is seen.

Discussion

Several reports have shown the feasibility and effectiveness of transcatheter occlusion of abnormal intrathoracic vessels with steel coils in children. The technique of embolization with mechanical devices was described in 1975.[1]

Embolization by interventional catheterization in children has now been reported for systemic-to-pulmonary collateral arteries,[2] pulmonary arteriovenous malformations,[4,5] surgically created Blalock-Taussig-shunts,[2] small patent arterial ductus,[6] coronary artery fistulas,[7-9] central venous connections[2] and arterial supply to pulmonary sequestrations.[2] As many as 100 coils have been delivered safely in the same young patient.[5] Other techniques of embolization have included detachable balloons,[4] gelfoam fragments,[4] bucrylate adhesive[10] and Ivalon particles.[11] Next to steel coils, detachable balloons have been most widely used. The advantage of coils as compared to detachable balloons is that they can be delivered through smaller catheters (usually 5F but as small as 3F). This is an important consideration in children.

Although early reports with small series of children treated successfully with steel coils date back to 1984,[3] the number reported so far is still small. In our series of 17 children, a high rate of success of embolization is shown. Thrombosis of the vessel at the site of embolization is usually expected to occur within 10 minutes.[12] At recatheterization after a median of 11 months after the intervention, six of the vessels with only subtotal occlusion as the immediate result were reassessed by angiography. This showed that total occlusion of the vessel had subsequently occurred in all. Thus, it can be assumed that subsequent thrombotic apposition can improve the initial result and complete the obstruction to blood flow.

The results of this study also show, that smaller vessels with a diameter of less than 4 mm can be treated with great success and a very small risk of complications such as migration of the coil. No failures or complications were encountered in this series when embolization was performed in these smaller vessels. Vessels with a larger diameter can still be treated with success, but failure of embolization and secondary migration of the coil after its release from the catheter were encountered only in larger vessels. This is not likely dependent on technical aspects as, in accordance with previous reports,[2] the helical diameter of the first coil to be delivered into a vessel was chosen so that it exceeded the inner diameter by 10-30%. One technical difference might be that, in the present study, the diameter of the vessel without distal stenosis as measured on angiographic film was considered relevant, and not the stretched diameter of the vessel after distal occlusion as proposed in a previous study.[2] This technical difference seemed of minor importance, as failure of embolization and secondary migration occurred equally distributed in vessels with and without distal narrowing. Of the 76 coils delivered, four devices (5%) migrated after release from the catheter. This complication was seen in vessels with the largest diameters in the whole series, although this finding did not reach statistical significance. Migration along the target vessel is the major complication. This complication can be due to improper selection of the coiled diameter of the device.[13] Increasing experience showed that this complication can be minimized by selecting coils with helical diameters exceeding the vessel diameter by 10-30%.[2,12] Consistent with the findings presented, secondary coil migration in about 4% of embolization attempts was observed in a recent series.[14] Dislocation seemed independent of the type of vessel occluded. Our results seem to indicate that migration of the coil is infrequent in smaller vessels but may be observed

more frequently when an embolization attempt is made in larger vessels (greater than 5 mm in this series). The recent introduction of controlled-release coils into clinical practice should further minimize the risk of secondary migration, since with this new technique, the coils can be released from the catheter once their proper position within the vessel has been verified. In the case of high flow vessels without stenosis, secondary migration can be further prevented by proximal balloon occlusion of the vessel to stop blood flow during coil deployement.

In the embolization of systemic-to-pulmonary collaterals, which is the main indication for this procedure in children, secondary migration occurs towards branch pulmonary arteries. In most instances, the coils can be safely retrieved with snares or basket-catheters.[14] In two of our patients, a dislocated coil was left in place in a branch of a pulmonary artery after angiography had proven unobstructed flow. Other authors have already demonstrated that dislocated coils can be left in place when flow is not obstructed.[2,14] In both of our patients with dislocated coils in a pulmonary artery, repeat catheterization at 8 and 11 months respectively confirmed persistent patency of those arteries. Nevertheless, an attempt of retrieval with baskets or 4F snaring catheters should be made in every case of secondary migration as it is not known what would be the long-term consequences of such foreign material in these localizations.

As has already been mentioned, children with complex congenital heart disease and systemic-to-pulmonary collateral arteries form by far the largest group of patients where embolization of intrathoracic vessels has to be considered. These aortopulmonary vessels can be divided into large, probably congenital collaterals, as typically observed for instance in tetralogy with pulmonary atresia, and into smaller and tortuous vessels which are probably acquired.[15] The physiologic significance of this second group of vessels, which have been observed in this series also after early corrective cardiac surgery (neonatal arterial switch operation for complete transposition), is poorly understood. Thus it becomes clear that guidelines to determine which of these vessels should be occluded (either at surgery or by interventional catheterization) are difficult to establish. Systemic-to-pulmonary collateral arteries have been reported to be associated with pulmonary bleeding and hemoptysis,[16] and rarely with congestive heart failure.[17] Embolization with coils allowed effective treatment of these complications.[17] Pediatric cardiologists for years have begun to occlude such vessels when observed at routine catheterization. In addition, a recent report showed that preoperative occlusion of such collaterals before a Fontan procedure might improve the post-operative course and avoid prolonged post-operative pleural effusions.[18]

One problem in occluding systemic-to-pulmonary collaterals is to determine whether vessels can be safely embolized without risking impaired pulmonary perfusion. Recently, pulmonary scintigraphy combined with occlusion using balloons has been recommended as a means of assessing such perfusion prior to embolization.[19] In our experience, selective angiography proved to be a sufficient evaluation prior to intervention. In none of the cases where the pulmonary circulation had been assessed in the follow-up after embolization of aortopulmonary collaterals was a pulmonary perfusion deficit detected.

In summary, transcatheter embolization in children of abnormal intrathoracic vessels by means of coils is effective and safe. It is an important therapeutic adjunct to surgical treatment in the pre- and postoperative course of children with complex cardiovascular malformations. Although complications are rare, the indication for this interventional procedure deserves careful evaluation.

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