Coil embolization of congenital thoracic vascular anomalies in infants and children

BRADLEY P. FUHRMAN, M.D., JOHN L. BASS, M.D., WILFRIDO CASTANEDA-ZUNIGA, M.D., KURT AMPLATZ, M.D., AND JAMES E. LOCK, M.D.

ABSTRACT When significant thoracic vascular anomalies occur in children, they may present surgical difficulties making operative management undesirable. The recent development of a new, accurate coil-delivery system has enabled us to embolize 17 vessels in five children by passing Gianturco steel coils coated with thrombogenic Dacron strands through No. 5 or 6 Fr end-hole catheters suitable for infants. Coils of 0.038 inch packed diameter were fed through the catheter lumen by a flexible guidewire emerging as 3, 5, or 8 mm diameter loose coils. Fifteen of 17 vessels were successfully occluded. No complications or errors in placement of coils occurred. Four of five children clearly benefitted from the procedure. One died in spite of partial occlusion. Coil embolization can be performed accurately and safely even in small infants with a high rate of successful occlusion and may prove to be a valuable adjunct to operative management.


ABNORMAL VASCULAR communications can occur within the chest either as isolated lesions or in association with other congenital anomalies. Their significance is quite varied; however, those that do warrant occlusion can present special surgical difficulties that make operative management undesirable.

Previous reports of nonoperative occlusion of thoracic vascular structures in children have described transcatheter embolization with an assortment of thrombogenic debris, including buckylate adhesive,1 detachable silicone balloons,2–5 and steel coils.6,7 In general, the use of these techniques has been restricted to older children and adults. Hazards have included inappropriate embolization5,6,8 of hardware with occlusions of normal vascular structures, as well as failure to occlude the vascular anomaly.2–4 In children, successful closure of only 10 thoracic vessels among 14 attempts has been described, with inadvertent embolization of two normal vessels.1–6

Thus experience in children with thoracic vascular anomalies is not sufficient to allow assessment of the risks and benefits of transcatheter embolization, nor does it permit comparison of the various techniques.

This report describes the steel coil embolization of 17 thoracic vessels in five children with a new, accurate coil-delivery system and compares results of that procedure with those of previously described techniques.

Methods

Five infants and children were shown at cardiac catheterization to have congenital thoracic vascular anomalies. Findings were reviewed in a combined cardiovascular surgery, vascular radiology, pediatric cardiology conference, and transcatheter embolization was recommended. Patients were recatheterized. Arterial or venous access was gained with a No. 5 or 6 Fr end-hole catheter, and embolization was accomplished with Gianturco steel coils, uniformly coated with thrombogenic Dacron strands (figure 1). As previously described,9 coils of 0.038 inch packed diameter were straightened over a thin wire and fed through the catheter lumen by a flexible guidewire, emerging in the abnormal vessel as a loose coil of 3, 5, or 8 mm diameter. Extremely flexible guidewires were used to avoid displacement of the catheter tip during the procedure. After placement of coils, occlusion occurred by thrombosis, generally within 10 min. If complete occlusion was not angiographically apparent 5 or 10 min after initial placement, smaller coils were lodged in the remaining vessel lumen to further obstruct the vessel. The first coil was chosen so that extruded diameter exceeded the angiographically estimated vessel lumen by about 50%.

Clinical reports

Patient 1. Our first patient was catheterized at the University of Minnesota Hospitals at 6 years of age and was shown to have multiple pulmonary artery-to-
pulmonary vein arteriovenous malformations. This child had severe cyanosis secondary to right-to-left intrapulmonary shunting and was severely polycythemic, with a hemoglobin of 24 g/dl. Because these malformations were both multiple and widespread, resection was not possible. She was instead treated by coil embolization of six lesions with some clinical improvement (figure 2). Over the ensuing 3 years, however, severe polycythemia recurred. She was recatheterized at 9 years of age, with a hemoglobin of 25 g/dl. Five additional arteriovenous malformations were identified and embolized. One that had been partially occluded at the first catheterization was completely occluded with a new coil. In all, 11 lesions were embolized, 10 of which were completely occluded and one of which was partially occluded. A twelfth vessel could not be entered with a catheter. Hemoglobin has been stable at 18 g/dl over the past year.

Patient 2. A second child, born with tetralogy of Fallot and pulmonary atresia, underwent a left Blalock-Taussig shunt at 3 years of age. She was subsequently shown to have four large bronchial arteries, two to each lung (figure 3). Before corrective operation when she was 12 years of age, tolerance of transient bronchial arterial occlusion was established with a balloon catheter. To minimize bronchial arterial blood flow to her lungs during cardiopulmonary bypass, three bronchial arteries were embolized with Gianturco coils. Occlusion of all three vessels was complete. One vessel that appeared partially open at the time of catheterization was documented at 6 month follow up to have been totally occluded. The fourth bronchial artery could not be entered for embolization.

Patient 3. A 7-month-old infant girl presented with cardiovascular collapse and disseminated intravascular coagulation from massive aorta–to–pulmonary vein shunting. A 5.5 mm diameter artery from the infradiaphragmatic descending aorta to the lower two-thirds of the right lung was identified and partially occluded with Gianturco coils. Occlusion remained incomplete at 1 month follow up. This infant died shortly thereafter of intractable pulmonary hypertension.

Patient 4. A 3-year-old child with double-outlet right ventricle, ventricular septal defect, and mitral atresia with a hypoplastic left ventricle had undergone a Blalock-Hanlon procedure and pulmonary arterial banding in infancy. Follow-up catheterization documented low pulmonary arterial pressure distal to the band site. When cyanosis later increased, a Glenn shunt was contemplated, but a persistent left superior vena cava was shown to communicate with the coronary sinus, left atrium, and paravertebral venous plex-

FIGURE 1. “Mini” Gianturco steel coil, uniformly coated with thrombogenic Dacron strands, suitable for transcatheter embolization through No. 5 or 6F end-hole catheters.

FIGURE 2. Angiograms from patient 1, with multiple pulmonary arteriovenous malformations. A. Injection of one malformation through a catheter traversing the pulmonary artery rapidly opacified the malformation (M) and its draining vein (V) but not the arterial feeder itself. B. After embolization, the branch of the pulmonary artery (PA) supplying the malformation opacified as well as arterial branches to normal lung parenchyma, but neither the malformation nor its draining vein are seen. A second, unrelated malformation (M2), which had previously been embolized, is superimposed on the draining vein in A but is visualized in B.
us (figure 4). Because such collaterals can cause a Glenn shunt to fail, the left superior vena cava was occluded with coils before creation of a Glenn shunt through a right thoracotomy. The Glenn shunt was well tolerated and resulted in a decrease in hemoglobin from 20 to 16 g/dl.

**Patient 5.** An infant was born with complex congenital heart disease, including tetralogy of Fallot, pulmonary atresia, parachute mitral valve, a large right bronchial artery to the right pulmonary artery, and a patent ductus arteriosus connecting an aberrant left subclavian artery to the left pulmonary artery (figure 5). Because of congestive heart failure, the right bronchial artery was surgically ligated. A 4 mm Gore-tex shunt was placed in an attempt to establish optimal pulmonary blood flow, and a Blalock-Hanlon atrial septectomy was performed.

Persistent congestive heart failure necessitated ductus occlusion, which was performed in this 3.5 kg, 5-month-old infant with severe inanition by passing three Gianturco coils through a No. 5F catheter introduced through the left axillary artery. One year later this child is home and has normal heart size, no evidence of heart failure, and a hemoglobin of 17 g/dl.

**Discussion**

We have treated five infants and children by steel coil embolization with good results and with no significant complications (table 1). Fifteen of 17 vessels entered were satisfactorily occluded, and four of five children clearly benefitted from the procedure. One died of pulmonary hypertension in spite of successful embolization and near total occlusion of the abnormal vessel.

These five patients represent four distinct indications for occlusion of congenital thoracic vascular anomalies. Embolizations were performed to eliminate right-to-left shunting of blood, to protect the lungs from excessive intraoperative bronchial blood flow, to eliminate left ventricular volume overload, and to avoid collateral blood flow subsequent to performance of a Glenn shunt.

In each patient, transcatheter embolization was performed in lieu of operative management. The patient with multiple pulmonary artery–to–pulmonary vein arteriovenous malformations could not be offered operative management because her malformations were multiple and widespread. In patient 2, bronchial arterial embolization was performed to facilitate surgical correction of tetralogy of Fallot. Bronchial arteries are not readily closed through a median sternotomy, and thoracotomy for bronchial occlusion necessitates delay of sternotomy for corrective operation. The infant with an aberrant artery connecting the infradiaphragmatic descending aorta to the right lung presented in shock and was a poor candidate for operation at the time of her initial cardiac catheterization. Embolization, although only partially successful, facilitated stabilization. Embolization of the persistent left superior vena cava before performance of a right sided Glenn shunt in patient 4 obviated the need for a separate left thoracotomy. Ductus occlusion was performed in patient 5 with tetralogy of Fallot and pulmonary atresia after balloon occlusion in the catheterization laboratory had documented adequacy of alternative sources of pulmonary blood flow, eliminating the need for reoperation in a child with severe inanition who was believed to carry a high operative risk.

In three of these patients, the vascular anomaly occluded was a component of complex congenital heart disease. In those patients, coil embolization was performed as an adjunct to operative management.

In both small infants, embolization was performed with a transarterial approach. Five French end-hole catheters were sufficient for coil placement. There was no evidence of compromised limb blood flow related to vascular access, although heparin was not administered.

Although not all vessels could be entered for embolization (17/19), it was highly successful when at-

**FIGURE 3.** Angiograms from patient 2, with tetralogy of Fallot and bronchial collateral arteries. A. Before embolization; B. after embolization of three vessels. Subtotal occlusion of one bronchial (arrow) was complete at 6 months follow up.
tempted (15/17). Unintentional embolization of normal vascular structures did not occur. Follow-up evaluation showed neither late migration of Gianturco coils nor recanalization of occluded vessels. Of the two occlusion failures, it may be pertinent that one occurred in a child with disseminated intravascular coagulation (patient 3). It is possible that because of this process, the coil lost thrombogenicity in situ, preventing complete thrombosis of the vessel even on resolution of the coagulopathy.

Other means of transcatheter embolization of thoracic vessels in children have been described. Bucrylate adhesive\(^1\) and detachable silicone balloons\(^2\text{-}5\) have also been used, but reports have been sporadic and do not permit definitive comparison to results with steel coils.

Bucrylate adhesive rapidly polymerizes on contacting blood to form a spongy plug but cannot be administered without first completely occluding the vessel to avoid distal embolization.\(^6\) Zuberbuhler et al.\(^1\) have used this technique to completely occlude two and partially occlude one bronchial artery in a 5-year-old child with tetralogy of Fallot.

Reports of the use of detachable silicone balloons in three children with pulmonary arteriovenous malformations,\(^2\text{-}3\) one child with bronchial arteries and tetralogy of Fallot,\(^4\) and one child with a persistently patent Blalock-Taussig shunt after repair of tetralogy of Fallot\(^5\) suggest that balloon embolization of thoracic vascular anomalies is feasible in children. It has not, however, been reported in an infant. Embolization with small balloons (1 mm collapsed and 4 mm distended) requires only a No. 4F catheter, but embolization with larger balloons (2 mm collapsed and 8 to 9 mm distended) requires a No. 9F end-hole catheter. Small balloons detached in arteriovenous malformations have been unintentionally embolized into normal vascular structures.\(^5\) During the procedure accidental detachment is a problem,\(^5\) and because the detachable balloon is directed by flow on exiting the introducer

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**FIGURE 4.** Angiograms from patient 4. A persistent left superior vena cava (A, arrow) connected to the coronary sinus, paravertebral venous plexus, and left atrium was occluded before a right-sided Glenn anastomosis (B).

**FIGURE 5.** A. Angiogram from patient 5, with tetralogy of Fallot, pulmonary atresia, a 4 mm Gore-tex shunt, and a long patent ductus arteriosus (arrow) connecting on aberrant left subclavian artery (LSCA) to the left pulmonary artery (LPA). B. Angiogram showing embolization of the ductus (arrow) performed to alleviate congestive heart failure.
TABLE 1
Results of coil embolization in infants and children

<table>
<thead>
<tr>
<th>Patient</th>
<th>Age/wt.</th>
<th>Vascular anomalies</th>
<th>Successful occlusionA</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>9 yr/24 kg</td>
<td>14 pul AV malformations</td>
<td>10/11</td>
</tr>
<tr>
<td>2</td>
<td>12 yr/27 kg</td>
<td>T of F and pul atresia, 4 bronchial arteries</td>
<td>3/3</td>
</tr>
<tr>
<td>3</td>
<td>7 mo/6.4 kg</td>
<td>1 systemic-pul AV malformation</td>
<td>0/1</td>
</tr>
<tr>
<td>4</td>
<td>3 yr/11.5 kg</td>
<td>Mitral atresia, persistent LSVC</td>
<td>1/1</td>
</tr>
<tr>
<td>5</td>
<td>5 mo/3.4 kg</td>
<td>T of F, pul atresia, PDA</td>
<td>1/1</td>
</tr>
<tr>
<td>Total</td>
<td></td>
<td></td>
<td>15/17</td>
</tr>
</tbody>
</table>

AV = arteriovenous; LSVC = left superior vena cava; PDA = patent ductus arteriosus; pul = pulmonary.

A No. of vessels totally occluded/No. of vessels embolized.

catheter, proper positioning can be difficult.4 Permanent vascular occlusion appears to occur over a period of 1 to 2 weeks with detachable balloons. In swine, balloons remaining inflated 10 or more days caused permanent occlusion of 91% of vessels.11 Changes in balloon volume caused by permeability of the balloon and late balloon deflation may prevent permanent occlusion.12 In all, four of six pulmonary arteriovenous malformations,2,3 two of three bronchial arteries,4 and the Blalock-Taussig shunt6 were successfully occluded with balloons.

Gianturco’s original steel coil device13 has been miniaturized14 for delivery through a No. 5F polyethylene catheter and has been used to embolize a Blalock-Taussig shunt in a child6 with inadvertent embolization of the pulmonary artery of the left lower lobe.

In the past, premature coiling of the device in the catheter has complicated coil delivery and impaired the accuracy of placement. However, Castaneda-Zuniga et al.9 have developed a method of coil delivery that greatly enhances accuracy of placement by preventing coiling in the catheter. By this technique, complications were not observed in our series. The 5F and 6F catheters sufficed, and the technique was safely ap-
plied to infants as well as to children. Successful occlusion was achieved in 15 of 17 vessels attempted.

Coil embolization appears to be safe and reliable, even in small infants. The procedure may be performed as an adjunct to the operative management of children with complex congenital heart disease and may provide a practical, cost-effective alternative to the operative management of certain congenital thoracic vascular anomalies.

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