Transcatheter Closure of Congenital Coronary Artery Fistulae: Immediate and Long-Term Follow-Up Results

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ABSTRACT

Background: Transcatheter closure of coronary artery fistulae (CAF) has emerged as an alternative to surgery, but there are no long-term outcome results.

Objective: We report immediate and long-term results in 24 patients after transcatheter closure of congenital CAF.

Methods: A total of 24 patients aged 5 to 56 years old with congenital CAF underwent attempted percutaneous transcatheter closure using various devices between November 1998 and August 2008. The immediate closure results and clinical follow-up were reviewed.

Results: Of the 24 patients, 20 (83.3%) underwent successful transcatheter closure using various devices. An angiogram right after device deployment revealed complete occlusion in 15 patients (75%) and trivial- to mild-residual flow in 5 patients (25%). Four patients (20%) had transient ST-T wave changes after the procedure. The left ventricular end-diastolic volume decreased from 165 ± 31.4 mm^3 to 128.6 ± 24.4 mm^3 ($P = 0.012$) 24 hours after procedure, and the cardiothoracic ratio from 0.57 ± 0.02 to 0.53 ± 0.01 ($P = 0.003$). Follow-up was 100% complete and ranged from 3 months to 10 years. There were no early or late deaths. All patients were asymptomatic with complete closure of CAF except 1 patient (5%) who had a recurrence of shunt at 6-month follow-up, which was re-closed by percutaneous technique.

Conclusion: Transcatheter closure of CAF is feasible and safe in anatomically suitable vessels and is a promising alternative to surgery in most patients.

Introduction

Primary coronary artery fistulae (CAF), the anomalous communication between a major subepicardial coronary artery and a cardiac chamber or thoracic vessel, are rare congenital anomalies.[1,2] Although most patients with such anomalies are asymptomatic, early treatment is recommended to prevent complications such as congestive heart failure, pulmonary hypertension, myocardial ischemia or infarction, fistula rupture with cardiac tamponade, and infective endocarditis.[3,4] Some authors even recommend closure of all congenital fistulae during infancy when patients are asymptomatic.[4] Traditionally, surgical correction has been the standard treatment. Although surgical treatment of coronary artery fistulas is safe and effective,[5] patients may be exposed to the risks of bleeding, postpericardiotomy syndrome, myocardial infarction, and recurrence.[6] Furthermore, surgery requires a median sternotomy and sometimes cardiopulmonary bypass. Transcatheter closure of CAF was first introduced in 1983[7] and has increasingly become the first line therapy in anatomically suitable cases. This technique has advantages over surgery that include: less cost, shorter recovery time and duration of hospitalization, and avoidance of thoracotomy and cardiopulmonary bypass. However, information on the long-term outcome after percutaneous CAF closure is lacking. The purpose of the present article was to evaluate the immediate and long-term follow-up results of transcatheter closure of CAF using various devices.

Methods

Patients

Between November 1998 and August 2008, 24 patients (14 females) with symptomatic CAF and coronary dilation (maximal coronary artery diameter > 5 mm) underwent transcatheter fistula closure, while those with additional complex cardiac defects requiring surgical management or with tiny CAF found incidentally during catheterization were excluded from the study. Written informed consent was obtained from all patients or their guardians before cardiac catheterization and angiography.

Devices

The occlusion devices used to close CAF in this series included: Amplatzer duct occluder (AGA Medical

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Corporation, Golden Valley, MN); detachable coil (PFM Company, Germany); Cook coil (Cook Cardiology, Bloomington, IN); Amplatzer ventricular septal occluder (AGA Medical Corporation, Golden Valley, MN); and Amplatzer vascular plug (AGA Medical Corporation, Golden Valley, MN). The devices and their delivery systems have been described in detail previously.[8–12] Device selection was based on the anatomic features of the fistula. Cook coils, which are delivered by using a 3 French microcatheter, were used primarily in smaller CAF, especially those with multiple small orifices at the drainage site. The detachable coils and plugs, both of which are deployed by using a guiding catheter, were used to close tortuous fistulae.

Procedure

Heparin was administered (100 units/kg) after the femoral venous and arterial accesses were established. After hemodynamic data were obtained, aortic root angiography and selective coronary angiography were performed to demonstrate the anatomy of the fistula, its drainage site, and identify distal coronary branches. Coronary angiograms were analyzed using a HICOR blood vessel analysis software QCA system (Siemens Quantcor, Siemens Medical Solutions, Forcheim, Germany) and the diameter of the fistula was measured. Feasibility of and approach to closure were determined by the number and location of drainage sites, the location of the proximal coronary branches, and the ability to cannulate the distal part of the fistula.

Device deployment was performed either anterogradely or retrogradely. The protocols of antegrade and retrograde deployment have been reported in detail previously.[3] Antegrade deployment was preferred in all patients who had a large fistula, while retrograde deployment was attempted when there was difficulty in establishing an arteriovenous wire loop through the fistula. Selective coronary angiography and aortic angiography was performed immediately after device deployment.

Follow-Up Protocol

Electrocardiogram (ECG), chest radiograph, and echocardiogram were performed 24 hours after the procedure. Every patient who underwent successful CAF closure was followed up clinically and by chest radiograph, ECG, and echocardiogram at 1, 3, 6, and 12 months during the first year and annually thereafter. Catheterization was reperformed when the above evaluations indicated. The demographic, hemodynamic, and follow-up data were encoded in a prospective database program (Microsoft Access 97, Microsoft, Redmond, WA).

Data Analysis

Data are expressed as a frequency or percentage for nominal variables and as mean ± standard deviation (SD) for continuous variables. Differences in parameters before and after the procedure were compared using a paired t test. A $P < 0.05$ value was considered statistically significant.

Results

Clinical Features of Patients

At the time of procedure, the mean patient age was 18.0 ± 13.1 years old and weight was 43.4 ± 21.2 kg. Details of the patients' characteristics are shown in the Table. Of these 24 patients, 15 (62.5%) were symptomatic, with dyspnea on exertion in 13 patients, chest pain in 3 patients, and heart failure in 1 patient. ECG showed left ventricular hypertrophy in 5 patients, ST-T changes in 3 patients, incomplete right bundle branch block in 2 patients, and left atrial enlargement in 1 patient. Echocardiograms at the time of admission revealed normal cardiac function in 23 patients. The mean left ventricular ejection fraction was 0.55 ± 0.02. Most of the CAF originated from the right coronary arteries and drained into the right heart (Table). Of the 24 patients, 2 had multiple fistulae, both of which originated from the left anterior descending coronary artery and the right coronary artery and drained into the pulmonary artery through multiple orifices.

Immediate Results of Device Closure

A total of 20 out of 24 patients (83.3%) underwent successful CAF closure. Devices were deployed using the transvenous approach in 8 patients (40%) and the transarterial approach in 12 patients. Details of approaches and devices in the transcatheter closure patient group are shown in Figure 1. In 4 patients, an arteriovenous wire loop was created, but failed to introduce the long sheath into the distal fistula from the femoral vein (Figure 2). These patients then underwent successful transarterial fistula closure. Closure using 1 device was attempted in all fistulae, but 1 CAF was closed by using 3 Cook coils due to its multiple small drainages. The mean diameter of the fistulae closed by using 1 device was 5.4 ± 1.7 mm and the mean size of the devices was 11.3 ± 0.7 mm. The fluoroscopy time was 39.8 ± 19.2 minutes and the procedure time was 145.8 ± 44.3 minutes. There were 4 patients who did not receive device closure in this series; 2 were due to multiple fistulae draining into the pulmonary artery through many small orifices and the other 2 patients had unsuccessful procedures due to extreme vessel tortuosity with inability to cannulate the distal fistula even using a percutaneous transluminal coronary angioplasty guide wire. These 4 patients eventually underwent surgical treatment.

Repeated angiograms following device deployment demonstrated complete CAF closure in 15 patients (75%). Two patients (10%) had trivial residual flow which disappeared 24 hours after the procedure demonstrated by checking the echocardiogram. A total of 3 patients (15%) who underwent detachable coil embolism had moderate

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Table 1. Anatomic Details of the Coronary Artery Fistulae

Patients	Gender	Age (y)	Weight (kg)	Fistula Origin	Fistula Drainage	Fistula Diameter (mm)
Intervention patients						
1	Female	6	20	RCA	RV	4.0
2	Female	7	21	RCA	RA	8.6
3	Female	7	18	LM	RA	6.0
4	Female	7	29	LAD	RV	5.0
5	Female	8	15	RCA	RV	4.2
6	Female	10	26	RCA	LV	5.7
7	Female	10	35	RCA	RV	3.6
8	Female	10	39	RCA	RV	5.0
9	Female	15	40	RCA	RV	4.5
10	Female	17	51	LCx	RV	5.0
11	Female	18	49	LAD	RV	2.2
12	Female	33	67	RCA	RV	5.4
13	Female	39	59	RCA	RA	4.0
14	Female	56	71	RCA	PA	MD
15	Male	5	20	RCA	RV	7.0
16	Male	15	55	LAD	RA	9.2
17	Male	21	75	RCA	RV	5.0
18	Male	22	75	RCA	LV	7.6
19	Male	29	81	RCA	RV	4.8
20	Male	33	51	RCA	RA	6.3
Non-intervention patients						
1	Male	34	51	LAD, RCA	PA	MD
2	Male	16	56	LAD, RCA	PA	MD
3	Male	5	16	LCx	RV	6.0
4	Male	9	21	RCA	LV	4.0

Abbreviations: LAD, left anterior descending coronary artery; LCx, left circumflex coronary artery; LM, left main coronary artery; LV, left ventricle; MD, multiple drainages; PA, pulmonary artery; RA, right atrium; RCA, right coronary artery; RV, right ventricle.

residual flow 15 minutes after device deployment. Supplementary Cook coils were added to each to reduce residual flow. One of them was completely closed and the other 2 had mild residual flow after procedure. There were 4 patients (20%) who had transient ST-T wave changes and slight increase in creatine kinase and cardiac troponin I, but only 1 had transient chest pain. There were no deaths, arrhythmias, strokes, infections, hemolysis, device migration, or fistula dissection.

Patient Follow-up

Follow-up was 100% complete and ranged from 3 months to 10 years (4.7 ± 3.2 years). An echocardiogram on the following day showed trivial residual flow in 2 patients. The mean left ventricular ejection fraction was 0.57 ± 0.02

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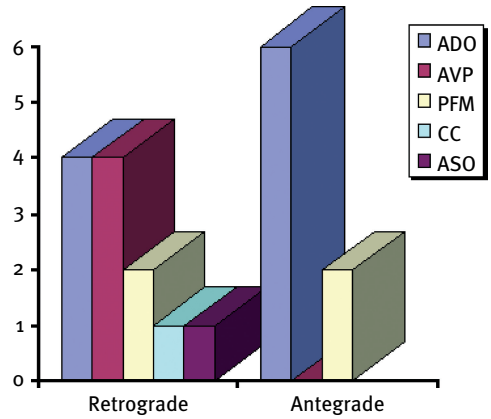

Figure 1. Devices and approaches in intervention patients. Abbreviations: ADO, amplatzer duct occluder; ASO, amplatzer ventricular septal occluder; AVP, amplatzer vascular plug; CC, cook coil; PFM, detachable coil.

and fractional shortening in the short axis view was 0.34 ± 0.01. The mean pulmonary artery pressure was 24.2 ± 2.2 mm Hg. Although there was no significant difference in the left ventricular ejection fraction and fractional shortening between before and 24 hours after procedure, the left ventricular end-diastolic volume decreased from $165.2 \pm 31.4\ mm^3$ to $128.6 \pm 24.4\ mm^3$ ($P = 0.012$) by echocardiography, and the cardiothoracic ratio from 0.57 ± 0.02 to 0.53 ± 0.01 ($P = 0.003$) by chest radiography. At the 1 and 3 month follow-up evaluation, all patients were in New York Heart Association functional class I with no clinically audible murmurs or residual flow. However, a continuous murmur was noted at the 6 month follow-up, and echocardiography revealed significant residual flow in 1 patient with CAF from the right coronary artery to the left ventricle which had been completely closed by using a 16-mm vascular plug. The patient was recatheterized. Coronary angiography showed the device in place and residual flow through the device (Figure 3). The fistula was completely closed without residual flow both angiographically and on follow-up echocardiography by using 4 Dacron stranded, stainless steel coils (Cook Cardiology, Bloomington, IN). At the last follow-up, all patients were asymptomatic. ECG showed no ST-T change and echocardiogram showed normal cardiac function with normal wall motion.

Discussion

Transcatheter CAF closure can be performed using various devices including detachable coils,[13] Amplatzer vascular plug,[12] Amplatzer duct occluder,[8] detachable balloons,[14] and covered stents,[15] while the vast majority of the fistulas were closed using microcoils.[10,16–18] Microcoils have made an important contribution to the technique of transcatheter CAF closure. The availability of 4 or 5 French end-hole steerable catheter facilitates access into the distal end of the tortuous fistula. Compared with the use of larger device such as the vascular plug and umbrella occluder, transcatheter coil embolization of CAF is relatively simple. Moreover, the flexible catheter delivery system is helpful for minimizing procedural complications such as puncture and dissecting of the fistula vessel and the use of coaxial system may provide better accuracy, stability, and precision for placement of microcoils.[17] However, many CAF have no adequate narrow drainage for coil delivery. In our experience, the Amplatzer duct occluder is an ideal device for CAF closure provided the drainage is large enough to allow the passage of the long sheath. This device can be deployed antegradely or retrogradely, and usually use of a single device is enough for complete closure. Another useful device for CAF closure is the Amplatzer vascular plug. This is a new occlusion device used for treating arteriovenous fistulae and other vascular malformations in the peripheral vasculature. They have a wide range of device sizes and can be delivered through a 5–8 French standard coronary guiding catheter because there is no fabric within the plug. Compared with umbrella devices, the plug affords greater opportunity to close the tortuous fistulae as it can be delivered through a flexible guiding catheter. Detachable coils can also be delivered through a guiding catheter, however, it seems that the plug has advantages over the detachable coil in ease of delivery and incidence of residual flow.

In order to reduce complications during the procedure, several procedural and clinical details mandate special attention. First, precise identification of the distal coronary branches is difficult before the fistula is closed due to fast blood flow through the fistula vessel. Thus, the optimal site for device placement is the fistula drainage. However, even after the device is placed at the fistula drainage, it does not mean that a small area of myocardial infarction would not occur. Therefore, it is necessary to monitor ST-T changes for 24 to 48 hours after CAF closure. Secondly, there are 2 approaches available for device deployment. The advantages of the transvenous approach over the transarterial approach are that the former avoids potential damage to the femoral artery and fistula vessel, allows use of a larger long sheath and a larger device, and offers an approach to close the tortuous fistula. However, this technique is unavailable for the fistulae draining into the pulmonary artery and the left heart. In our experience, the transarterial approach is as safe as the transvenous approach provided the fistula is large enough to pass the long sheath through it. In this series, apart from Cook coils, which can be delivered using a 3 French microcatheter, and detachable coils and vascular plug, which are delivered using a guiding catheter, we have successfully implanted 5 umbrella devices into CAF retrogradely without major cardiac complications. Thirdly, the sizing of the device is difficult due to variation of the fistula morphology. We chose a device with a waist size, which was roughly double to the diameter of the fistula drainage in accordance with the sizing of the device for

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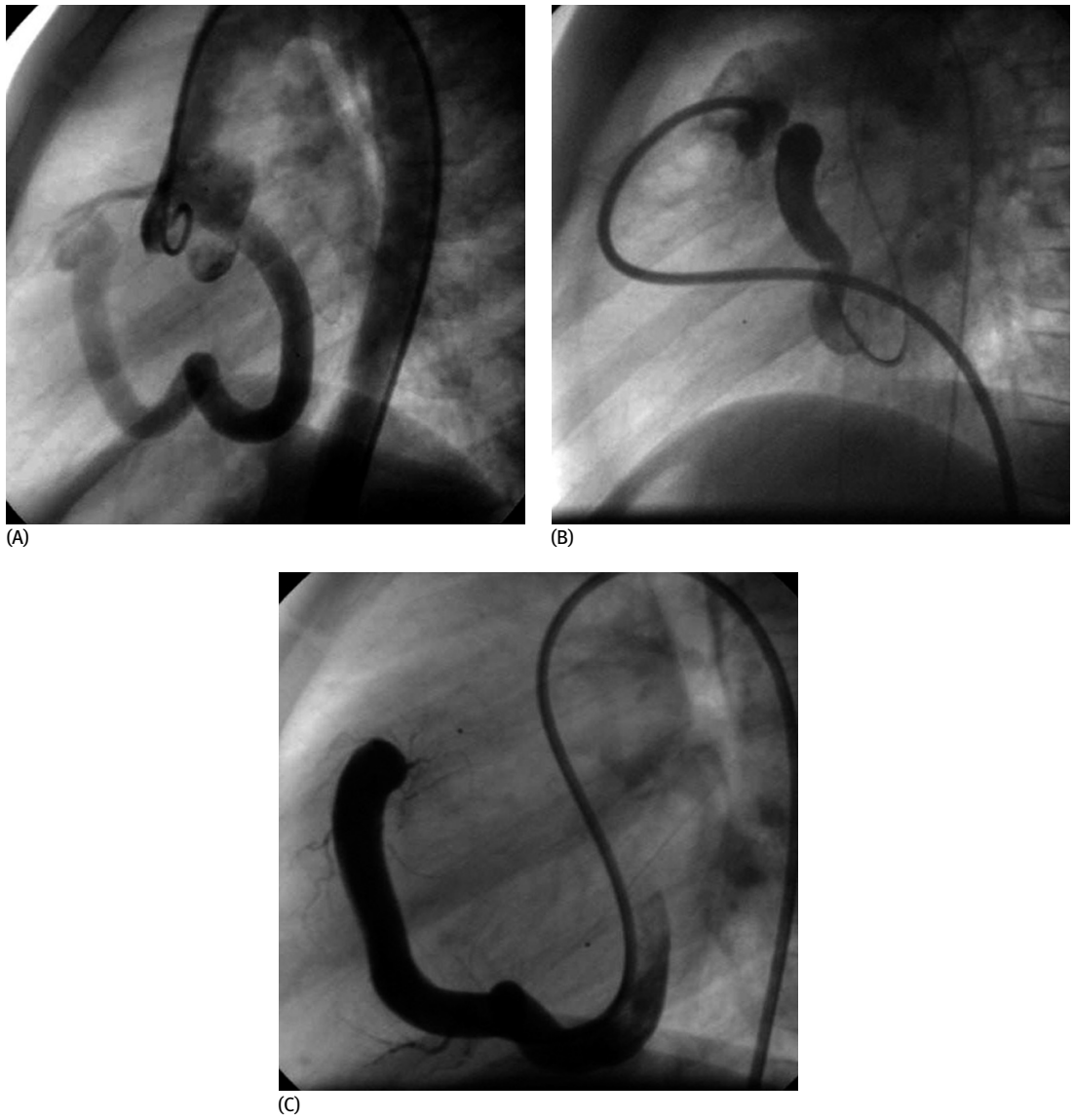

Figure 2. Transcatheter closure of a tortuous fistula from femoral arterial approach. (A) Pre-closure. (B) An arteriovenous wire loop failing to enable passage of the long sheath into the distal fistula. (C) Complete closure following transarterial placement of a device.

closure of tubular patent ductus arteriosus since most of the fistulae looks tubular at the drainages. Undersizing of the device may lead to complications such as residual shunt and device migration, especially when the patient is a child.

Several reports have evaluated the efficacy and safety of transcatheter closure of CAF.[15–18] The procedural complications included transient ischemic changes, unretrieved device embolization, fistula dissection, myocardial infarction, and transient atrial arrhythmia in the literature and there was 1 procedure related death. So far only 1 report has evaluated the midterm outcome of transcatheter closure of CAF, where all 4 patients underwent closure using microcoils.[10] In our series of 24 patients, 20 underwent successful transcatheter CAF closure using various devices without major complication. During follow-up, all patients were asymptomatic except 1 who had recanalization of the fistula.

Conclusion

Transcatheter closure of CAF is safe and feasible in anatomically suitable cases with ideal long-term outcome.

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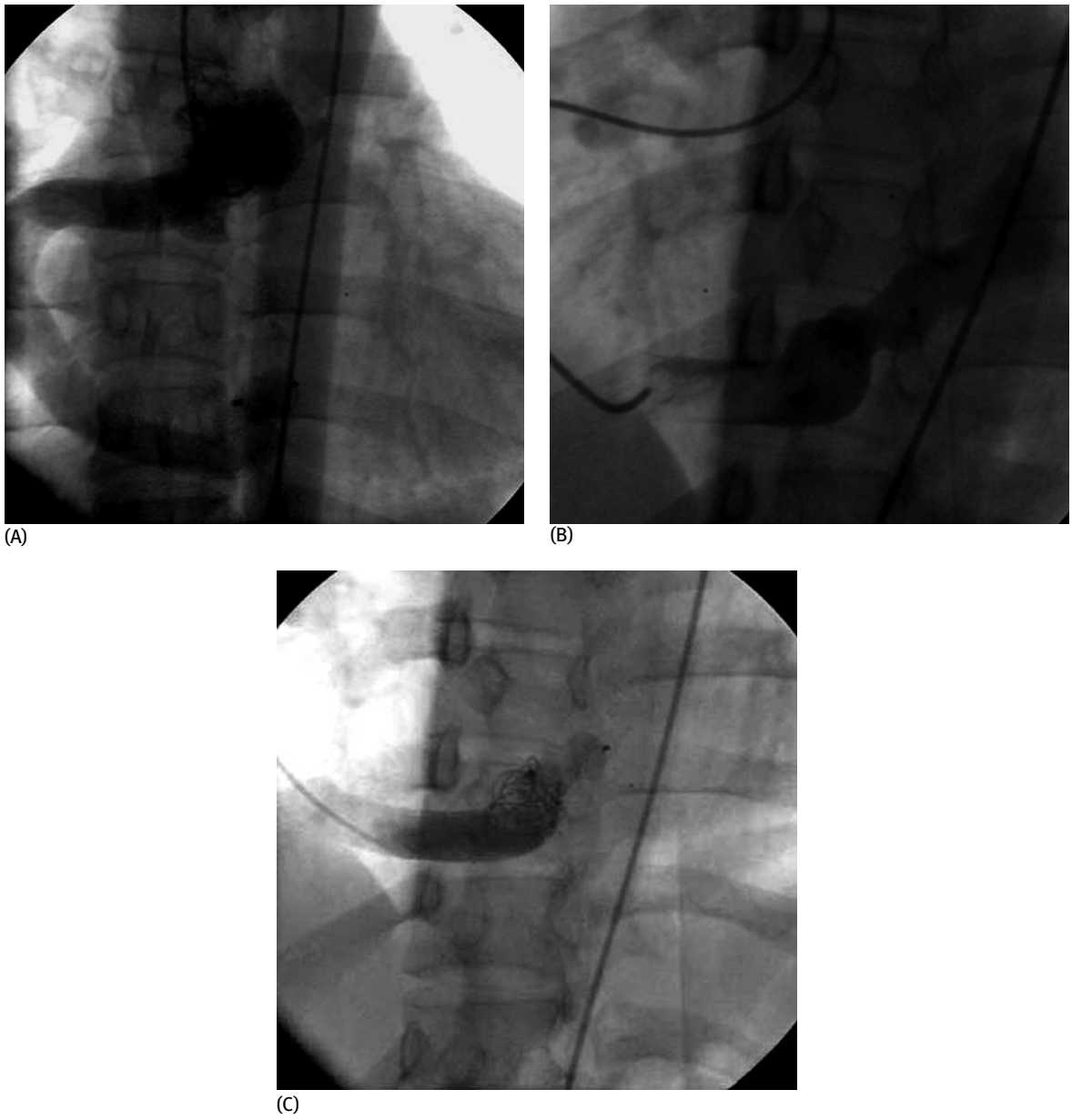

Figure 3. Recanalization of fistula 6 months after device closure. (A) Check angiogram showing complete closure. (B) Angiogram showing the device in place and residual flow through the device. (C) Complete closure of the shunt after coil embolization.

Complications such as residual shunt and myocardial infarction may occur within 6 months after procedure. The device selection and delivery technique should be based on the anatomic characteristics of the fistula.

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