Uneventful Follow-Up 2 Years after Endovascular Treatment of a High-Flow Iatrogenic Aortocaval Fistula Causing Pulmonary Hypertension and Right Heart Failure

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ABSTRACT: Iatrogenic aortocaval fistula is an extremely rare pathologic condition that often results in clinically significant left-to-right extracardiac shunt. In slow-progressing cases, chronic right-sided heart failure can occur and, in some patients, may persist for years. We present a patient with a long-standing aortocaval fistula that was causing high-flow left-to-right shunting, tricuspid regurgitation, severe pulmonary hypertension, and right-side heart failure. After undergoing complete endoscopic isolation of the aortocaval fistula, the patient experienced dramatic clinical improvement and continued to have excellent imaging and clinical resolution after 2 years of follow-up.

INTRODUCTION

Aortocaval fistula is a rare cause of clinically significant extracardiac left-to-right shunt. Up to 80% of cases described in the literature result from rupture of the aortic aneurysm, with 15% resulting from penetrating trauma.[1] James Syme described this pathologic condition for the first time in 1831,[2] Cooley conducted the first successful open-surgery repair in 1955,[3] and the first stent-graft exclusion was made in 1998.[4] We report a case of aortocaval fistula causing right-heart failure that was successfully treated with two-step endovascular embolization.

CASE PRESENTATION

A 59-year-old male was admitted in our hospital for clinical evaluation and diagnostics. He had experienced symptoms of isolated right-sided heart failure for the last 10 years—mainly dyspnea and fatigue during common physical activity and edema in the lower extremities. He also reported symptoms of pulsating abdominal formation over the past few years and had spontaneous audible pulsatile bruit around his body. His reported medical history was remarkable with right nephrectomy 25 years earlier due to benign tumor formation as well as controlled arterial hypertension and permanent atrial fibrillation.

During clinical examination, we observed a large palpable periumbilical abdominal formation, hepatomegaly, and pretibial edema. There was also an extremely loud abdominal systolic-diastolic murmur with maximum intensity at the right abdominal quadrant. An electrocardiogram showed normofrequent atrial fibrillation with an incomplete right bundle branch block and no repolarization abnormalities. Transthoracic echocardiography revealed normal left ventricular dimensions and left ventricular ejection fraction (62%), biatrial dilatation more pronounced on the right side (left atrial area 28 cm^2, right atrial area 35 cm^2), and right ventricular dilatation and hypertrophy (basal diameter 51 mm, free wall thickness 8 mm). He had light mitral regurgitation and severe tricuspid regurgitation with an estimated right ventricular systolic pressure of 90 mm Hg. His inferior vena cava was extremely dilated (62 mm) in the whole abdominal segment, with Doppler characteristics of artery-like blood flow.

Contrast-enhanced CT aortography revealed a missing right kidney and an enlarged remnant of the right renal artery with an s-shaped course in the axial plan. There was also communication between the right renal artery and inferior vena cava (IVC). The IVC had a maximal width of 72 mm with dense contrasting of the vein in the arterial phase of the study and a high-flow left-to-right shunt (Figure 1).

Our multidisciplinary cardiovascular team discussed various treatment strategies in this case. Classic open surgery was deemed inappropriate due to extremely high perioperative predictive mortality, mostly because of the bleeding risk. Endovascular stent graft implantation was discussed as an option but was not covered by the patient's healthcare insurance. Another option was coil embolization.

The first reported endovascular treatment was performed by Cekirge et al. on a patient with an aortocaval fistula after nephrectomy in 1996, just 2 years before the first stent graft exclusion procedure.[5] Cekirge's team used 39 coils followed by injection of rapid polymerizing glue. Based on the literature, we discussed a stepwise approach to avoid a sudden interruption of the left-to-right shunt, which carries the risk of extreme acute hypotension because of the acute decrease in venous return.

Figure 1.
Initial computed tomography angiography shows a missing right kidney, enlarged remnant of the right renal artery with s-shaped course, and a huge communication to the inferior vena cava that contrasted intensely during the arterial phase due to a high-flow left-to-right shunt.

As a first step, we implanted two devices—a left atrial appendage (LAA) and patent ductus arteriosus (PDA) occluder (Occlutech International, AB)—in the ostium of the right renal artery (Figure 2). This caused a moderate decrease of the fistula's flow. On contrast CT performed the next day, migration of the ostial device was established, and high-flow arteriovenous (AV) communication was restored with no significant difference in IVC contrast during the arterial phase (Figure 3). The two devices were stuck on the thinnest and most tortuous part of the AV communication, which allowed us to use them during a second procedure as an anchor for initial basket formation and "packaging" with coils.

Control computed tomography angiography (CTA) a day after the first endovascular procedure showed dislodgment and distal migration of the previously implanted PDA occluder caused by the high flow into the fistula. Note in Figure 2 that the arterial phase contrasting of the IVC is restored.

During a second endovascular procedure that was performed 1 month later, 13 embolization Penumbra Ruby coils (Penumbra, Inc.) were implanted using the conglomerate LAA/PDA occluder as anchoring and stabilization to avoid distal migration of the first several coils used to build the initial basket (Figure 4). We have used a 6F catheter with right radial vascular access (which is much more anatomically appropriate due to the acute angle at the target vessel's origin), a 6F multipurpose guiding catheter, and a Trailblazer microcatheter (Medtronic) for super-selective cannulation of the AV communication.

At the end of the procedure, there was a significant reduction in blood flow (Figure 4) and a 2-fold drop in pulmonary artery pressure (from 92 mm Hg to 50 mm Hg). One month after the second procedure, an echocardiogram showed

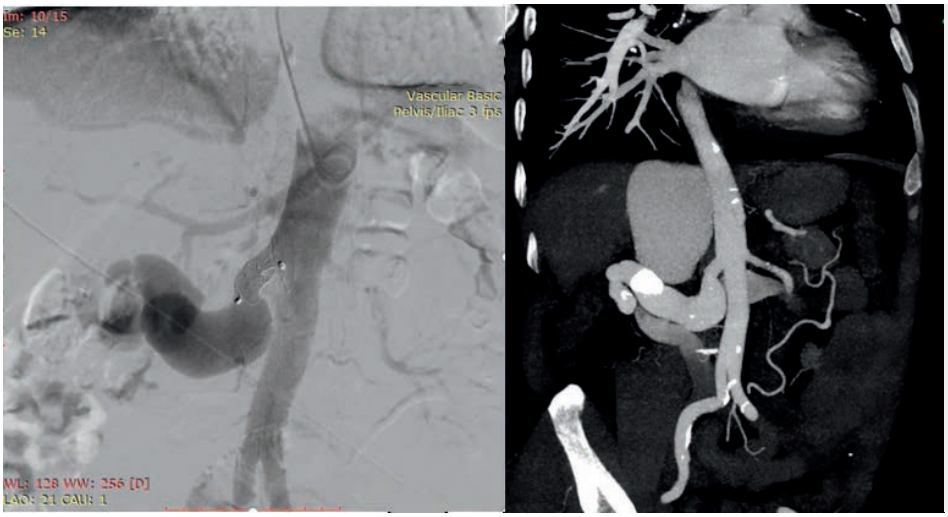

Figure 2.
Left: The final ostial position of the Occlutech PDA Occluder (Occlutech International) with the inferior vena cava contrasted later in relation to the arterial phase. Right: Control computed tomography, which was performed on the next day.

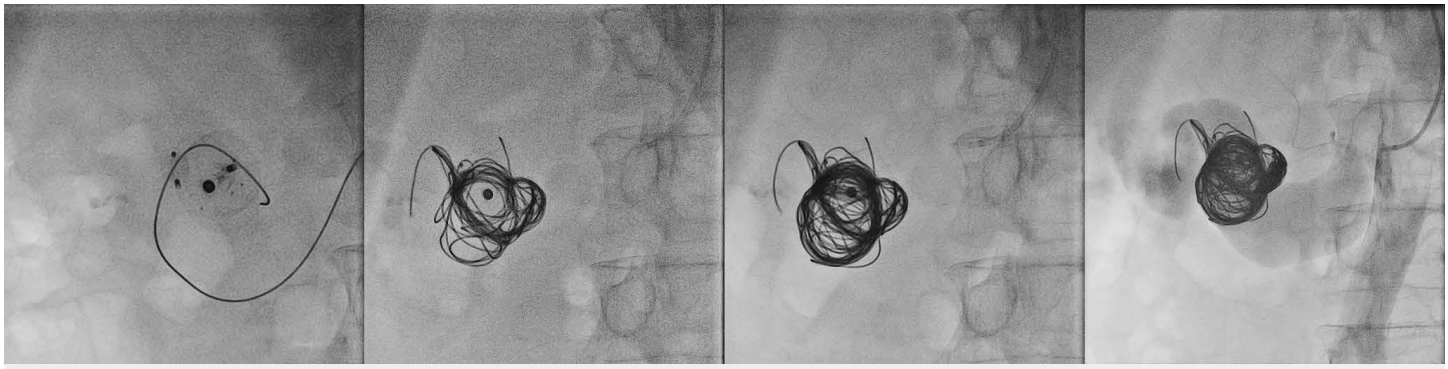

Figure 3.
Stepwise embolization with coils of the arteriovenous communication using the stable position of the patent ductus arteriosus/left atrial appendage occluders for an anchoring maneuver to avoid distal coil migration. Note the attenuation of the contrast intensity in the inferior vena cava.

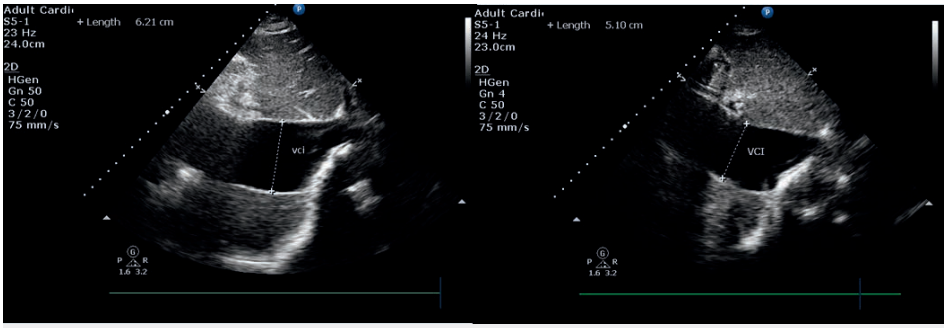

Figure 4.
Initial (left) and 1-month follow-up (right) echocardiograms show pronounced regression of the inferior vena cava maximal diameter from 62.1 mm to 51 mm.

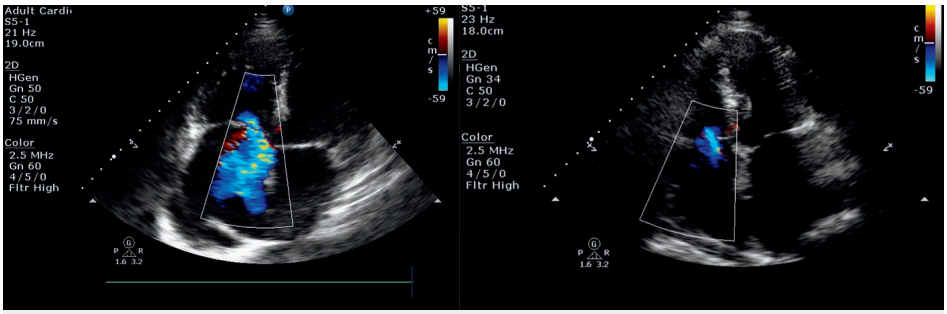

Figure 5.
Initial (left) and 1-month follow-up (right) echocardiograms show an impressive reduction of the third-degree tricuspid regurgitation to "trace" and reduction of the right atrium.

a reduction in IVC size and a further reduction in tricuspid regurgitation (Figure 5).

The 2-year follow-up was uneventful, with the patient showing a complete lack of symptoms and complete resolution of the murmur compared to his NYHA functional class III at baseline. On physical examination, the palpable formation and systolic-diastolic murmur were not detectable, and his tricuspid regurgitation had improved from "severe" to "trace." We also observed a significant drop in estimated pulmonary artery pressure, from 92 mm Hg at baseline to 40 mm Hg. Pleural effusion was completely resolved. The control CTA (Figure 6) showed complete isolation of the aortocaval fistula with no contrast in the IVC during the arterial phase. All aortic branches except the right renal artery were patent, and IVC size was reduced by half.

DISCUSSION

Aortocaval fistula is a rare, life-threatening, pathologic condition. An iatrogenic cause is extremely uncommon, with only a handful of successfully treated

Figure 6.
Comparison between the initial (left) and 24-month follow-up (center) computed tomography angiography and follow-up computed tomography (right) shows stable position of the embolization materials, complete isolation of the aortocaval communication, and full interruption of the left-to-right shunt. The green star marks the inferior vena cava.

cases found in a literature search. Although surgical repair has been performed successfully, it is still associated with a high mortality rate. However, in select patients, endovascular treatment is an acceptable alternative.

Despite the lack of treatment guidelines for aortocaval fistula, stent-graft implantation has the most published experience. Unfortunately, it has been associated with late endoleak in some cases. Like the Cekirge case described in the literature, stepwise embolization for a high-flow AV fistula is a viable and efficient option. The upper (radial) remote vascular approach can be considered as a method of choice for the majority of elective visceral and renal endovascular interventions because of the acute angle of arterial origin. The applied strategy has the advantages of being a low-risk, minimally invasive approach with no need for general anesthesia and a short hospital stay.

CONCLUSION

Peripheral iatrogenic AV communication is a rare cause of pulmonary hypertension and right-sided heart failure. Endovascular treatment has become an effective alternative for treating high-flow AV communications in select patients. In the case described, a two-stage embolization procedure using an initial anchor with big occluder devices and additional filling coils resulted in a safe and efficient treatment.

Conflict of Interest Disclosure:
The authors have completed and submitted the *Methodist DeBakey Cardiovascular Journal* Conflict of Interest Statement and none were reported.

Keywords:
iatrogenic aortocaval fistula, left-to-right shunt, arteriovenous communication

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