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Coil Embolization of Spontaneous Splenic Arteriovenous Fistula for Treatment of Portal Hypertension

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Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
Funds Collection G

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Conflict of interest: None declared

Patient: Female, 64
Final Diagnosis: Splenic arteriovenous fistula
Symptoms: Left lower quadrant abdominal pain
Medication: —
Clinical Procedure: Coiling embolization
Specialty: Surgery

Objective: Rare disease

Background: Splenic arteriovenous fistula (AVF) is a rare cause of portal hypertension which may manifest with abdominal pain, diarrhea, ascites, and/or hematemesis. Fistula formation may be traumatic or spontaneous. Eighty-six percent of spontaneous splenic AVFs occur in women, and 55% are associated with a preexisting splenic artery aneurysm.

Case Report: A 64-year-old Caucasian female with unremarkable past medical history presented with new onset of left lower quadrant abdominal pain and persistent diarrhea. CTA demonstrated dilated mesenteric veins consistent with portal hypertension. A 1-cm splenic artery aneurysm associated with a splenic AVF was identified and confirmed by celiac angiography.

The splenic artery was embolized both distal and proximal to and within the aneurysm sac. Completion arteriography showed minimal flow throughout the splenic artery, and there was no flow into the splenic AVF.

Conclusions: Traditionally, splenectomy has been the definitive treatment, but coil embolization has been recently reported. Successful coil embolization of a splenic AVF is described. Physicians should be aware of this pathology as an etiology of portal hypertension.

MeSH Keywords: Embolization, Therapeutic • Hypertension, Portal • Splenic Artery

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Background

Splenic arteriovenous fistula (AVF) is a rare cause of portal hypertension, often presenting with symptoms of abdominal pain, diarrhea, ascites, and/or hematemesis [1]. The pathology may be identified following traumatic or inflammatory injury or may occur spontaneously [1]. The first reported cases in the 19th century were identified at autopsy [2,3]. For most of the 20th century, the diagnosis was made during exploratory laparotomy and later by diagnostic arteriography. Currently, computerized tomographic angiography (CTA) offers a less invasive means of diagnosis. Most patients have been successfully treated by surgical ligation or splenectomy. However, coil embolization has recently been reported with excellent results [4]. A case of spontaneous splenic AVF is reported, which was managed successfully by coil embolization. The patient has consented to the description of her case for this report. Physicians should be aware of this uncommon etiology of portal hypertension, which may be cured with a minimally invasive approach.

Case Report

A 64-year-old Caucasian female with an unremarkable past medical history presented with new onset of left lower quadrant abdominal pain and persistent diarrhea. The physical examination was unremarkable, including a soft, nontender abdomen without audible bruit. Preoperative liver function studies and hemoglobin were normal. CTA demonstrated

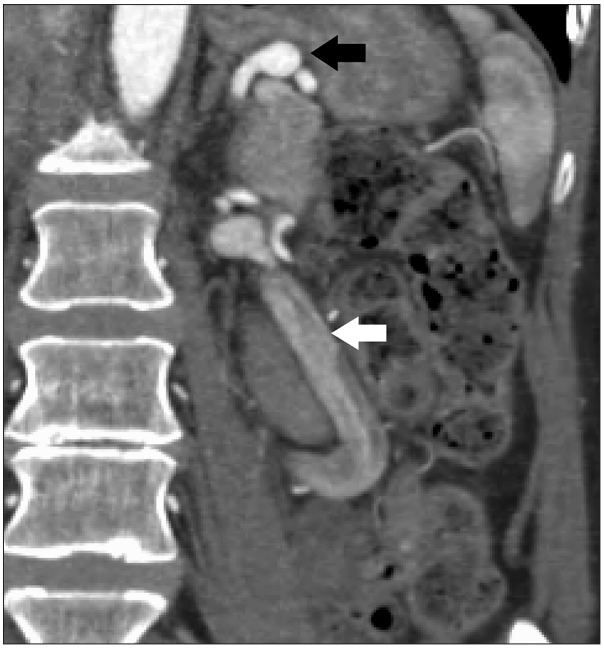

Figure 1. Splenic artery aneurysm (black arrow) and distended inferior mesenteric vein (white arrow).

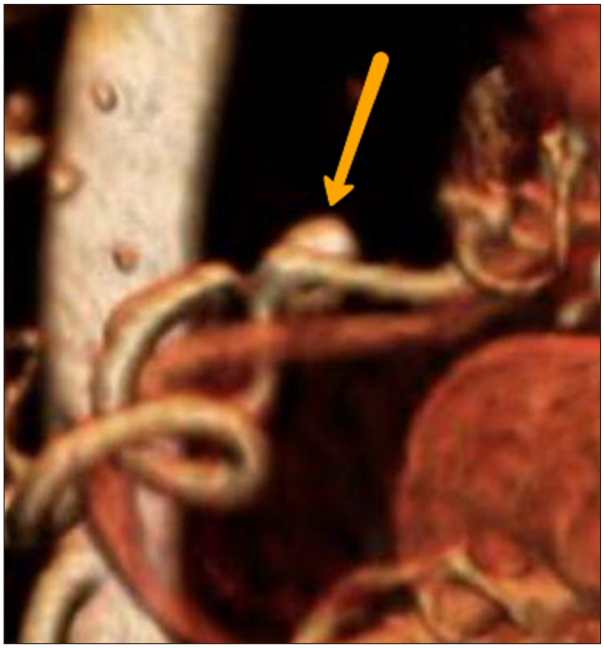

Figure 2. Splenic artery aneurysm (orange arrow).

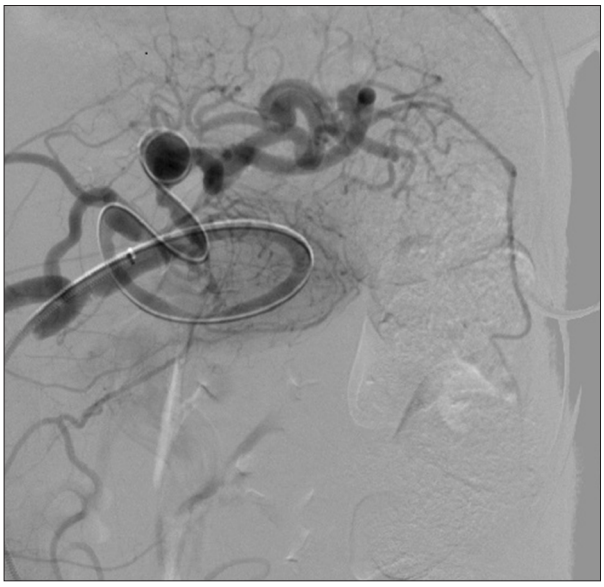

Figure 3. Coil embolization of splenic AV fistula.

dilated mesenteric veins consistent with portal hypertension, although there was no evidence of splenomegaly or ascites. A 1-cm splenic artery aneurysm associated with a splenic AVF (Figures 1, 2) was identified and confirmed by celiac angiography. Contrast was immediately apparent, communicating directly from the splenic artery aneurysm to the splenic vein.

The splenic artery aneurysm and AVF were accessed percutaneously through a femoral artery approach with a 6 French guiding sheath and occluded by coil embolization with Terumo (Somerset, NJ) HydroCoils without complication (Figure 3). The

Table 1. Summary of literature review: Splenic AV Fistula.

Etiology	Traumatic	Spontaneous
	29 (34%)	56 (66%)
Treatment	Surgical splenectomy or ligation	Surgical splenectomy
	14 (48%)	47 (84%)
	Coil embolization	Coil embolization
	14 (48%)	5 (9%)
Sex	Male	Male
	15 (52%)	8 (14%) 50% with splenic artery aneurysm
	Female	Female
	14 (48%)	48 (86%) 56% with splenic artery aneurysm

splenic artery was embolized both distal and proximal to and within the aneurysm sac. Completion arteriography showed minimal flow throughout the splenic artery, and there was no flow into the splenic AVF. Surveillance CT scans at 6 and 12 months post-procedure confirmed complete occlusion of the aneurysm and AVF. There was no evidence of splenic infarction. The patient remains asymptomatic 3 years post-procedure.

Discussion

A literature search was conducted for English language reports of splenic AVF over the last 30 years. The bibliography of each publication was then inspected for additional reports. Publications in a language other than English were excluded from this review. Cases of intra-parenchymal splenic arteriovenous connection were also excluded. Seventy-three publications were identified describing 85 patients with an average age of 45 years, 73% of them female.

Twenty-nine patients (34%) with splenic AVF were associated with prior trauma, upper abdominal surgery, or pancreatitis [1,5–32]. One of these patients suffered a splenic arterial mycotic embolus associated with valvular endocarditis, which created the splenic AVF [32]. Of this group, 15 (52%) were male, representing 65% of the total male group. The average age of the male patients was 34.7 years, which was significantly less than the average age of female patients (50.6 years) associated with traumatic origin (P<.020). Fourteen (48%) of the traumatic patients were successfully managed by coil embolization. One patient received no treatment. The remaining 14 patients underwent surgical splenectomy or ligation, with a 7.1% mortality rate.

Fifty-six patients (66%) developed a spontaneous splenic AVF [1,33–73], of which 31 (55%) were associated with a splenic artery aneurysm ranging in size from 1 to 15 cm in diameter. Within this spontaneous origin group, 48 patients (86%) were female, with an average age of 48 years, and only 8 patients (14%) were male, a significant difference (P<.001). Twenty-seven females (56%) were found to have a splenic artery aneurysm associated with the AVF. Four men (50%) had a splenic artery aneurysm associated with the AVF; 47 patients (84%) were treated with splenectomy with a 4.25% mortality rate; 5 patients (9%) were successfully treated with coil embolization (Table 1); 2 patients were identified at autopsy, and 2 patients were not treated.

Although no patients with splenic AVF managed by endovascular coil embolization were reported to suffer a splenic infarction, splenic infarction has been reported rarely after coil embolization for splenic artery aneurysm. Presumably, blood supply through the short gastric arteries is sufficient to preserve the spleen in most cases. Clearly, embolization of a distal splenic aneurysm or AVF could result in a wedge-shaped partial splenic infarction. Splenic infarction can often be managed nonoperatively, although removal may be necessary for persistent pain or evidence of infection.

Conclusions

Splenic AVF is a curable etiology of portal hypertension. Although splenectomy can be performed with acceptable mortality, coil embolization may now be considered a first line of therapy, with no reported mortality, as long as arterial access is possible. Physicians should be aware of this pathology as an etiology of portal hypertension.

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