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Case Report

Endovascular management of incidentally discovered splenic arteriovenous fistula resulting from ruptured splenic aneurysm: Case report and review of the literature ☆

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ABSTRACT

Splenic arteriovenous fistulas (SAVFs) are rare vascular anomalies, which have a described association with splenic artery aneurysms. Treatment options include surgical fistula excision, splenectomy, or percutaneous embolization. Here we present a unique case of endovascular repair of a splenic arteriovenous fistula (SAVFs) associated with a splenic aneurysm. A patient with past medical history of early-stage invasive lobular carcinoma was referred to our interventional radiology practice to discuss an incidentally discovered splenic "vascular malformation" discovered during magnetic resonance imaging of the abdomen and pelvis. Arteriography demonstrated smooth dilatation of the splenic artery, with a fusiform aneurysm which had fistulized to the splenic vein. There were high flows and early filling of the portal venous system. The splenic artery, immediately proximal to the aneurysm sac, was catheterized using a microsystem and embolized using coils and N-butyl cyanoacrylate. Complete occlusion of the aneurysm and resolution of the fistulous connection was achieved. The patient was discharged home the following day, without complication. Associated splenic artery aneurysms and SAVFs are rare occurrences. Timely management is necessary to prevent adverse sequelae such as aneurysm rupture, further enlargement of the aneurysmal sac, or portal hypertension. Endovascular treatment, including n-Butyl Cyanoacrylate glue and coils, offers a minimally invasive treatment option, with facile recovery and low morbidity.

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Introduction

Splenic arteriovenous fistulas (SAVFs) are rare vascular anomalies with a described association with splenic artery aneurysms. Commonly described causes of SAVFs include trauma, inflammatory injury, and idiopathic occurrence [1]. It is thought that spontaneous rupture of a splenic artery into the vein can cause SAVFs, with women of childbearing age and a history of multiparity being most at risk [2,3]. In 2017, Garrett et al. performed a 30-year literature review of SAVFs and found 73 publications describing 85 patients, the majority of which were female. Spontaneous SAVFs occurred in 66% of patients and 55% of those were associated with a splenic artery aneurysm. Surgical splenectomy was the most common treatment of SAVFs, utilized in 84% of cases, with an associated 4.25% mortality rate [4].

Treatment of SAVFs is important to prevent portal hypertension and its sequelae. Treatment options include surgical fistula excision, splenectomy, or percutaneous embolization. Recent data support positive outcomes associated with endovascular (EV) treatment of splenic vascular anomalies, which allow for splenic preservation with low associated morbidity [5,6]. We present a unique case of EV repair of a SAVF associated with a splenic aneurysm using coils and n-Butyl Cyanoacrylate glue.

Case report

A 49-year-old female was referred to our interventional radiology (IR) practice by her oncologist. She was diagnosed with early stage (T1c pN0), invasive lobular breast carcinoma 2 years prior and was treated with lumpectomy and axillary node dissection and radiation therapy; she is on maintenance leuprolide (Lupron, AbbVie, North Chicago, IL) given every 4 weeks. During her initial work-up, a screening magnetic resonance imaging (MRI) of the abdomen and pelvis was obtained, which revealed several liver hemangiomas and a, "Probably benign splenic lesion." Subsequent MRI reports detailed a splenic vascular malformation, which was largely unchanged in size. After 32 months of relative stability, the patient was referred to IR to discuss management of the vascular malformation. In anticipation of the IR consult, a triphasic computed tomographic angiography (CTA) was ordered.

The patient was seen virtually in our IR clinic by one of the interventional radiologists. Her medical history was significant for breast cancer and Raynaud's. She had no history of trauma, including motor vehicle accidents, and no history of abdominal surgeries. She had a total of 5 pregnancies, without reported complications. The patient's cross-sectional imaging was reviewed. There was dilation of the splenic artery, which measured up to 7 mm in diameter, and the splenic vein, which measured up to 11 mm in diameter. At the splenic hilum there was a fusiform, aneurysmal dilatation, thought to be arterial in origin. The density of contrast on CT was near-identical on the arterial and delayed phases in the splenic artery and vein. Within the splenic parenchyma there was a dilated, tangle of vessels (Fig. 1). Notably, there were no imaging findings of portal hypertension, such as splenomegaly, hepatomegaly, ascites, or varices. The patient had mild thrombocytopenia, platelets preprocedure measured 129, but no history of gastrointestinal bleeding.

The patient was scheduled for treatment one week after her clinic consultation. The patient was consented for arteriography and possible embolization, with plan to treat if there was a lesion amenable to EV repair. The procedure was performed under general anesthesia and right common femoral access was obtained. The celiac artery was selected with a Simmons 1 catheter (Cook Medical, Bloomington, IN) and arteriography was performed. There was preferential flow in the splenic artery relative to the common hepatic artery and the splenic artery was diffusely enlarged. At the splenic

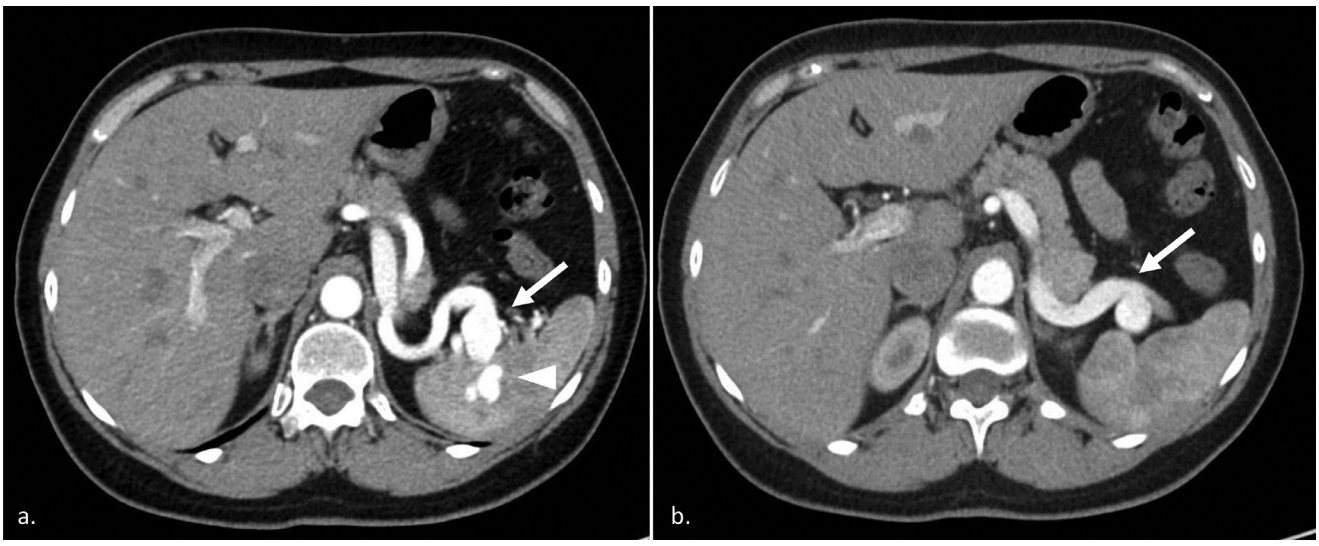

Fig. 1 – CT of the abdomen in arterial phase demonstrates fusiform arterial aneurysm (arrow) at the splenic hilum with intrasplenic tangle of vessels (arrowhead) (A). Figure (B) demonstrates dilated splenic vein (arrow).

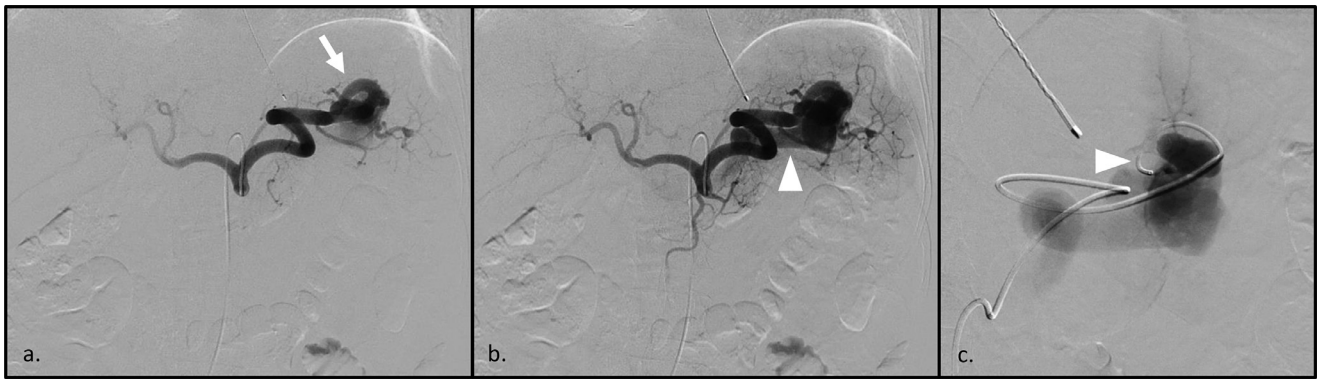

Fig. 2 – Angiography of the celiac axis demonstrates dilated aneurysm of the hypertrophied splenic artery (arrow) with early filling of the splenic vein (arrowhead) (A and B). Selective catheterization of the terminal aspect of the artery associated with the splenic arteriovenous fistula (arrowhead) was performed for embolization (C).

hilum there was an aneurysmal outpouching and rapid filling of the splenic and portal veins (Fig. 2). The Simmons-1 catheter was exchanged for a 4 Fr glide Cobra (Terumo, Somerset, NJ), which was advanced into the distal splenic artery, immediately proximal to the vascular abnormality. Selective splenic arteriography demonstrated early filling of the splenic vein and portal vein along with an abnormal tangle of vessels within the spleen parenchyma. The neck of the aneurysm was selected with a 2.8 Fr Progreat catheter (Terumo, Somerset, NJ). An anchor coil, 2 mm x 4 cm AzurCX detachable coil (Terumo, Somerset, NJ), was placed in a nearby artery. The splenic artery, immediately proximal to the fistula, was embolized with one 7 mm x 6 cm micronester coil (Cook Medical, Bloomington, IN) and three 7 mm x 24 cm AzurCX detachable coils (Terumo, Somerset, NJ). Postembolization angiography demonstrated sluggish filling of the splenic vein, at which point the decision was made to embolize with n-Butyl Cyanoacrylate glue (TRUFILL n-BCA Liquid Embolic System, Cordis, Miami Lakes, FL). The glue dilution consisted of 1 milliliter (mL) of n-Butyl Cyanoacrylate glue mixed with 2 mL of ethiodized oil (Lipiodol Ultra Fluid (UF), Guerbet, Princeton, NJ). Completion angiogram demonstrated complete stasis of the treated splenic artery, supplying approximately 30% of the splenic parenchyma (Fig. 3). The patient was discharged home the following day, without complication. One month after angiography and embolization the patient presented for routine follow-up with a CTA. The patient had returned to her baseline health, without any pain. Her CTA revealed a small splenic infarct in the upper pole, her splenic artery and vein were normal in caliber, and coils were seen at the site of the aneurysm (Fig. 4). She has had no further medical care or imaging within our system related to her splenic AVF.

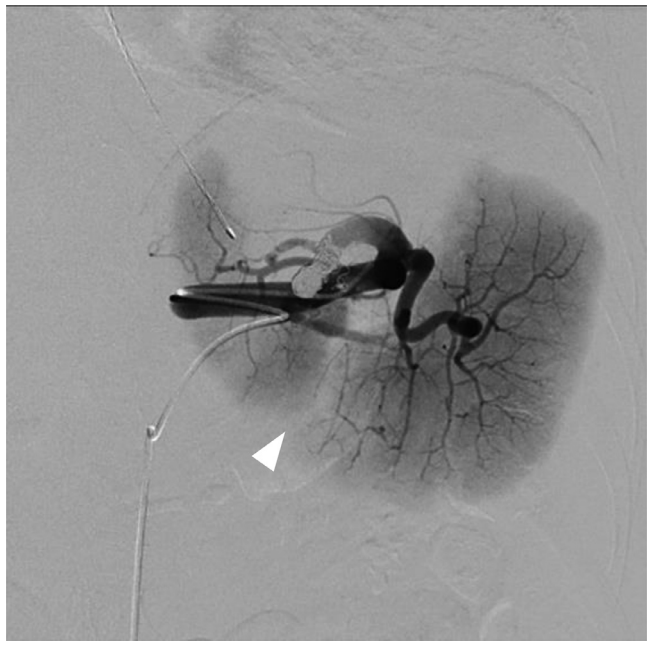

Fig. 3 – Angiography of the splenic artery after coil and n-Butyl Cyanoacrylate glue embolization demonstrates complete occlusion of the splenic aneurysm and fistula. There is a small parenchymal defect, in keeping with devascularized splenic parenchyma (arrowhead).

Discussion

SAVFs are rare occurrences, with variable treatment options described in the literature. Patients, such as ours, can be asymptomatic at the time of presentation; however, SAVFs can lead to rapid onset portal hypertension, with associated gastrointestinal bleeds, diarrhea, ascites, or high output heart failure. Timely management is necessary to prevent adverse sequelae. Historically, surgical treatment of splenic vascular anomalies has been standard of care; however, SAVFs are technically challenging to repair due to the oft-distal location of lesion, formation of adhesions, and portal collaterals [2].

Most of the available literature comparing open and EV repair has focused on treatment of splenic artery aneurysms with coil embolization. A large systematic review describing outcomes of splenic artery aneurysms treated with open surgical repair, EV repair, and conservative management

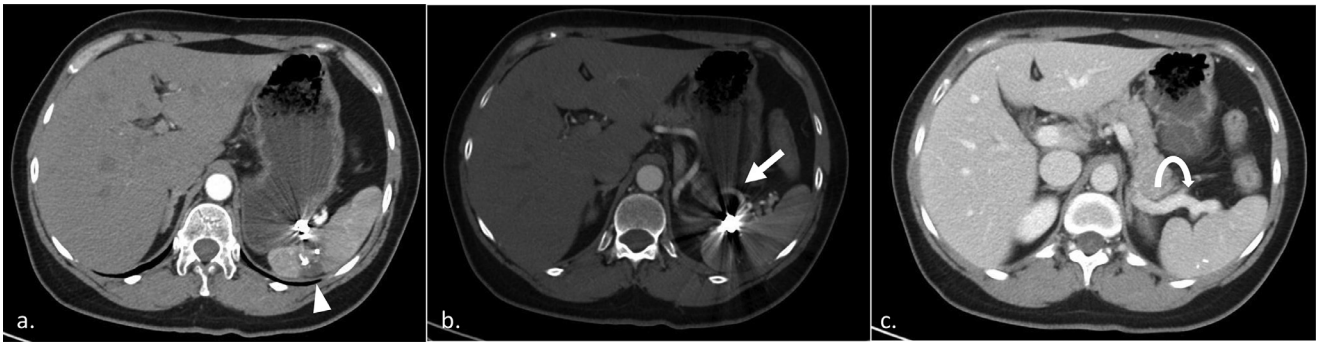

Fig. 4 – CT of the abdomen in arterial phase demonstrates a small infarct of the spleen (arrowhead) (A). The splenic artery (arrow) and vein (curved arrow) have decreased in size after coiling (B and C).

demonstrated similar early technical success between the open and EV treatment, although EV repairs were associated with a higher rate of reintervention. Of note, the 30-day mortality was significantly higher in the open repair compared to the EV group [6]. The higher reintervention rate associated with EV treatment of splenic artery aneurysms is likely secondary to vessel recanalization seen with mechanical embolic agents.

The available literature of EV treatment of SAVFs describes treatment with coil embolization [2,4,7–10]. Our case represents the first SAVF in the literature treated with both coil embolization and n-Butyl Cyanoacrylate glue. The coils served as a scaffold and produced a low arterial flow state in the diseased aspect of the splenic artery, and the glue resulted in near instantaneous occlusion of the SAVF. The authors' theory was that glue would provide a more durable treatment response, with lower rates of recanalization, due to the longer-lasting occlusion seen with cyanoacrylates [11,12]. The available literature describing EV repair of SAVFs has not described occurrence of splenic infarct [4]. In our patient, a small upper pole infarct was identified on 1 month follow-up imaging. The distal, selective nature of our case and utilization n-Butyl Cyanoacrylate glue of may have attributed to this finding. Notably, the patient was asymptomatic, and no intervention was required. Infarction and ischemia are known complications associated with use of n-Butyl Cyanoacrylate glue [13]. Other rarer complications described when using n-Butyl Cyanoacrylate glue include nontarget vascular embolization, paradoxical cerebrovascular or pulmonary emboli have been associated with variceal embolization, and sepsis [14–17].

Conclusion

Endovascular therapies offer a minimally invasive, safe treatment option for SAVFs, with several case series in the literature describing good technical success with no incidences of mortality described. Utilization of n-Butyl Cyanoacrylate glue and coils, proves to be a safe embolic combination with theoretical benefits of lower recanalization and reintervention rates.

Patient consent

Informed consent was obtained from all individual participants included in this case report.

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