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Giant Splenic Aneurysm with Arteriovenous (A-V) Shunt, Portal Hypertension, and Ascites

Authors' Contribution:
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: Male, 43
Final Diagnosis: Splenic aneurysm
Symptoms: Ascites • fever • portal hypertension
Medication: —
Clinical Procedure: —
Specialty: Surgery

Objective: Rare co-existance of disease or pathology

Background: Splenic aneurysms are rare, asymptomatic, and usually derive from previous surgical interventions. Endovascular repair is the best option, but when A-V shunt is present, open repair might be more suitable.

Case Report: A 43-year-old man presented to the Internal Medicine Department of AHEPA University Hospital with symptoms of fever and ascites. He was an ex-medical student with a history of sickle cell anemia, who had undergone urgent splenectomy and cholecystectomy 26 years ago and had a transit ischemic attack at the age of 21 years. Diagnostic imaging control revealed a giant splenic aneurysm 9.8 cm in diameter and 5 cm in length, with a concomitant A-V shunt (due to common ligation of the vessels after splenectomy and long stump presence with concomitant erosion of arterial wall). The patient underwent open surgery and cross-clamping the orifice of the splenic artery, also including the splenic vein, and the vessels were ligated. Post-operatively, the patient remained in the Intensive Care Unit for 48 h and suffered a portal vein thrombosis treated with appropriate anticoagulants. One month later, he had acute hemorrhagic pancreatitis and paralytic ileus and underwent laparotomy performed by general surgeons.

Conclusions: Giant splenic aneurysms are rare and are usually caused by previous splenectomy and preservation of a long-vessel stump. Immediate surgical repair is mandatory because of the high risk of rupture.

MeSH Keywords: Aneurysm, Ruptured • Arteriovenous Fistula • Ascites • Conversion to Open Surgery • Hypertension, Portal • Splenic Diseases

Abbreviations: **ICU** – Intensive Care Unit; **A-V shunt** – arteriovenous shunt; **H.I.T.** – heparin-induced thrombocytopenia; **DSA** – digital subtraction angiography; **VAC** – vacuum-assisted closure; **CT scan** – computed tomography scan; **CT-angio** – computed tomography angiography; **Hct** – hematocrit

Full-text PDF: https://www.amjcaserep.com/abstract/index/idArt/911106

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Background

An aneurysm is an abnormal dilation of a vessel to more than 50% of its normal diameter. The splenic artery has a normal diameter of 0.46 cm, and the aneurysm diameter range is 1–2 cm [1]. Giant splenic aneurysms have diameters over 9.5 cm and are rare. A diameter over 2 cm has increased risk of rupture and requires surgical repair [1,2].

Splenic aneurysms are the most frequent visceral aneurysms (60%), followed by abdominal and iliac aneurisms, and their overall incidence in the general population is about 0.8% [1]. They are true aneurysms and they are 4 times more frequent in women than in men (4: 1). Pregnancy is a common risk factor related to rupture and they are often seen in 5th and 6th decades of life [1].

The vast majority (80–97.5%) are asymptomatic. Some patients, however, present non-specific symptoms such as epigastric or left upper-abdominal quadrant pain (49%), nausea, vomiting, digestive disorders, and anorexia. Non-specific symptoms are often related to giant aneurysms due to their expansion in the intraperitoneal cavity, occupying space and compressing adjacent organs. The main life-threatening complication is rupture (28%), which is accompanied by acute symptoms such as sudden onset of sharp abdominal pain, Kehr sign, gastrointestinal bleeding, hemoperitoneum, and hypovolemic shock [1–3].

The most common causes of acquired splenic aneurysms are surgical abdominal interventions, including splenectomy, and vessel ligation with a long stump [1] (Table 1). Splenic aneurysms are usually treated endovascularly [2,3], but when an arteriovenous shunt is present, open repair might be appropriate. In our case, endovascular repair was not an option because the huge dimensions of the A-V shunt would lead to systematic iatrogenic embolization and acute hepatic necrosis.

The aim of this article is to present the management of an extremely rare case of a giant splenic aneurysm with concomitant arteriovenous shunt, portal hypertension, ascites, paralytic ileus, and peritoneal infection due to hemorrhagic pancreatitis. In the current literature, only 9 similar cases are universally registered [4–9].

Case Report

A 43-year-old, man presented at the Internal Medicine Department of Ahepa University Hospital with symptoms of fever and ascites. He was an ex-medical student with an anamnesis of sickle cell anemia, who underwent splenectomy and cholecystectomy on urgent basis 26 years ago and had a transit ischemic attack at the age of 21 that forced him to abandon his medical studies. A diagnostic imaging control with CT scan was performed, revealing a large amount of free ascites and a giant mass in the left upper-abdominal quadrant, paraaortic and underneath the pancreas (Figure 1). A following CT-angio with i.v. contrast infusion in arterial and venous phases was performed, revealing a giant splenic aneurysm 9.8 cm in diameter and 5 cm in length, located in the previous common ligation of the splenic vessels (Figures 1–3). The most interesting finding was the concomitant contrast infusion into the enlarged splenic vein (an AV-shunt, due to common ligation of the vessels after splenectomy and long stump presence) (Figure 3).

Table 1. Splenic aneurysm and AV-shunt causes.

Splenic aneurysms	Splenic arteriovenous shunt
Congenital	Congenital
Acquired	Acquired
Portal hypertension	Rupture of splenic artery aneurysm into the splenic vein
Atherosclerosis	Penetrating trauma
Inflammation (e.g., pancreatitis)	Post-splenectomy, gastrectomy
Pregnancy	Mycotic infection
Abdominal trauma	Pancreatitis
Arterial degeneration (e.g., medial fibrodysplasia)	Iatrogenic
Collagen vascular disease	
Autoimmune disease (e.g., systematic lupus erythematosus, polyarteritis nodosa)	
After liver transplant	

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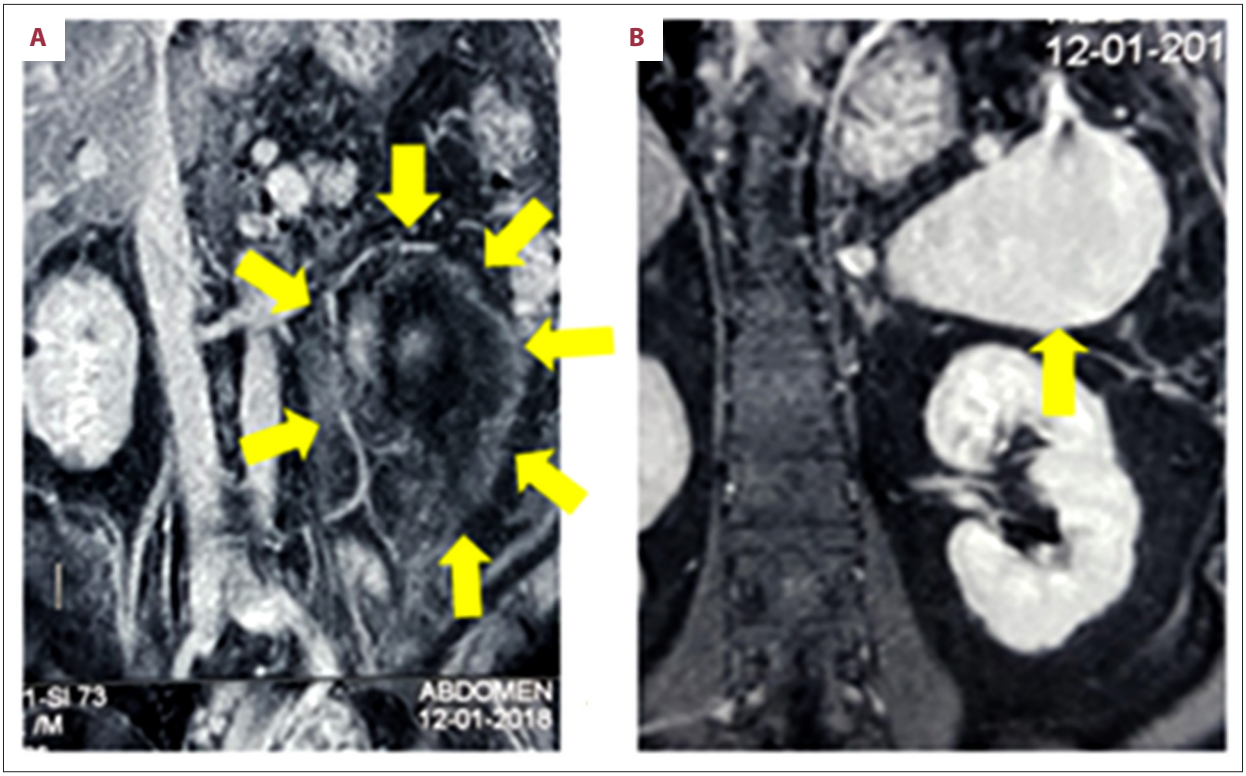

Figure 1. (**A**) CT scan: The arrows show the giant mass on the left upper abdominal quadrant. (**B**) CT-angio: The arrow shows the giant splenic aneurysm.

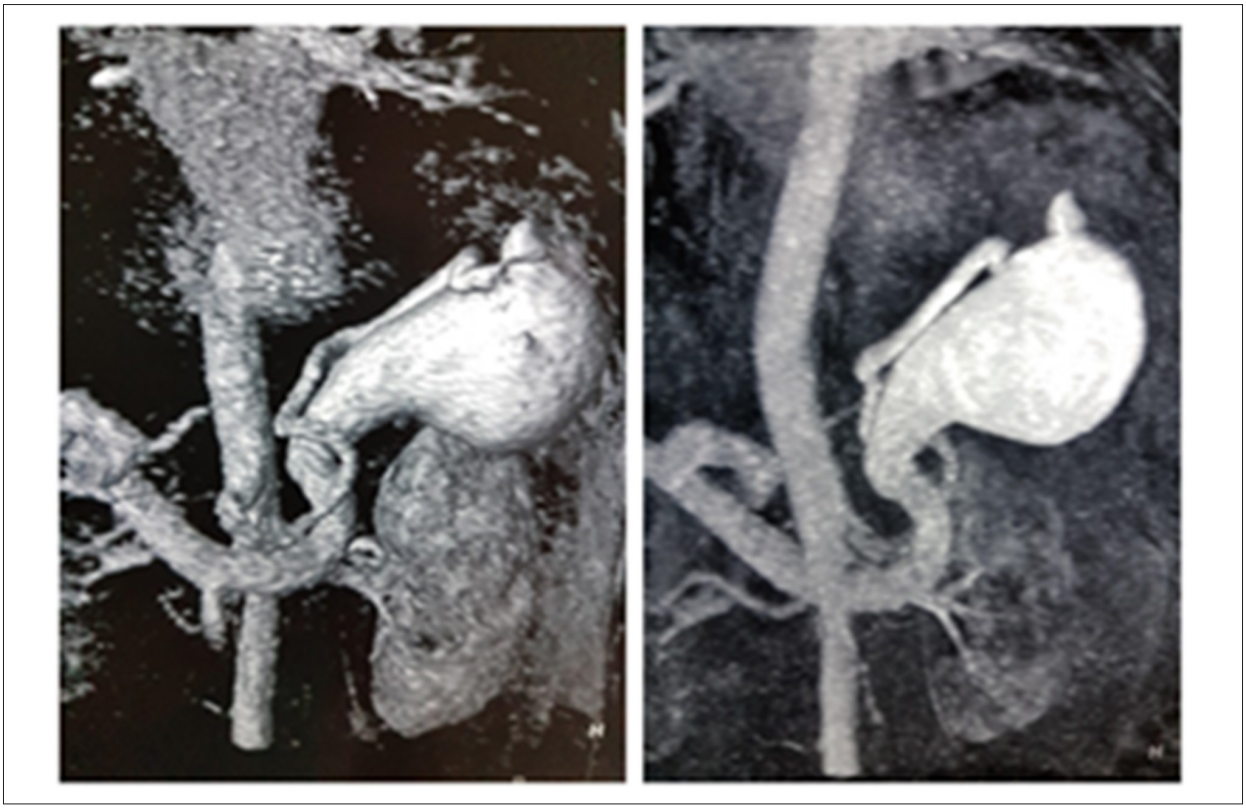

Figure 2. Splenic artery aneurysm and AVF imaging: CT-angio reconstruction with i.v. contrast infusion.

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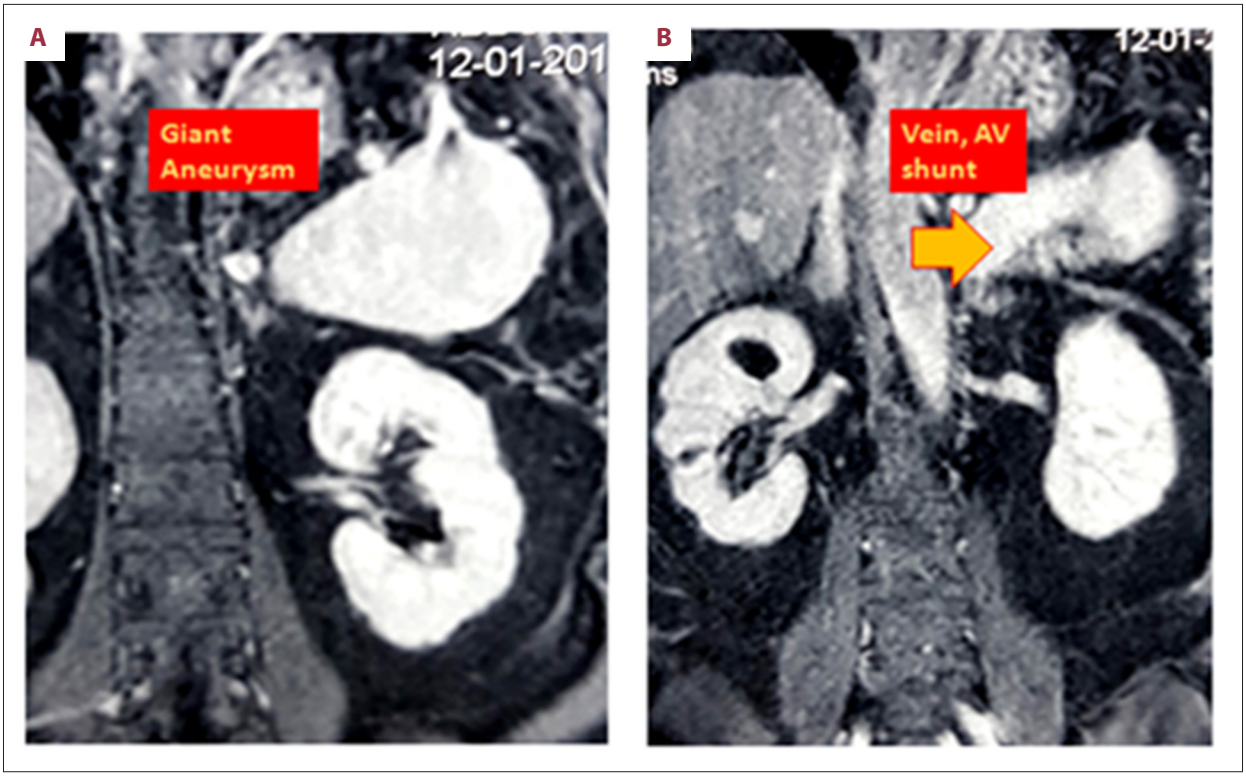

Figure 3. CT-angio. (**A**) Arterial phase, (**B**) Venous phase: The arrow shows the contrast into the enlarged splenic vein.

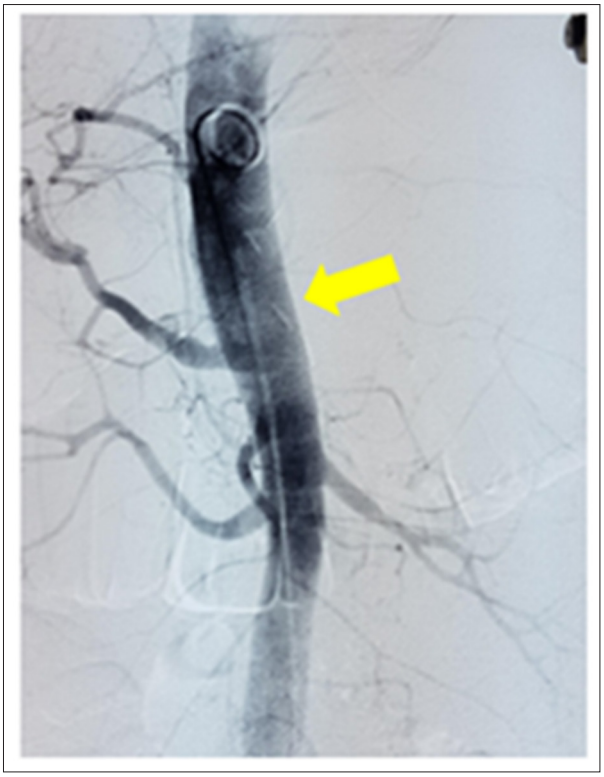

Figure 4. Post-operative DSA. a) Splenic artery orifice is not depicted (ligated para-aortically-arrow), b) Splenic vein is not depicted (ligated), c) A-V shunt is not depicted, d) Free contrast inside peritoneal cavity is not depicted (no major/minor bleeding).

A diagnostic centesis was performed by internists to exclude malignancy, and the patient was transferred to the Vascular Surgery Clinic. His long-term pharmaceutical treatment was aspirin (100 mg×1), hydroxyurea (500 mg×2), and tazobactam 4.5 g×4 (i.v.), and omeprazole 40 mg×1 (i.v.), spironolactone 25 mg×1 (p.o.), rifaximin 200 mg×3 (p.o.), saccharomyces boulardii 2×3 (p.o.), and enoxaparin 0.4 ml×1 (s.c.) were added preoperatively. Twenty-four hours later, he underwent surgery. An open repair via supra-infraumbilical incision was performed. Intraperitoneal access to the aorta was obtained via dislocation of the colon and small intestine. The lesion was included into a giant omentum sac, and was extremely fragile and hemorrhagic due to the chronic inflammatory process, and a covered rupture was revealed (Figure 4). We found 4 liters of ascites and blood in the peritoneal cavity. Sac preparation was extremely difficult due to the hemorrhagic and inflammatory tendency of the patient. The omentum and the aneurysmatic sac were prepared and removed from the left paracolic space (Figure 5). The upper half of the sac was in full contact with the pancreas, and remained unprepared to avoid organ lesion. A cross clamp to the orifice of the splenic artery including the splenic vein

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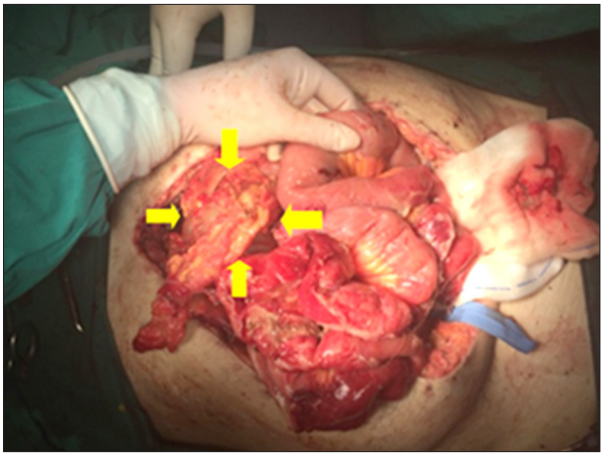

Figure 5. The arrows show the omentum sac of the aneurysm.

was performed and the vessels were ligated using 4-0 Prolene sutures and Teflon implants. Post-operatively, the patient remained in the ICU for 48 h and then was transferred to the General Surgery Department. Three days after the operation, he suffered a portal vein thrombosis and his antithrombotic treatment was modified from a prophylactic dose of Tinzaparin (3.500 IU anti-Xa per day) to a therapeutic dose of Fondaparinux (to avoid H.I.T., 7.5 mg×1 per day). On the 10th post-op, day he was transferred to the Internal Medicine Department. Six days later, he developed a fever with concomitant high levels of laboratory inflammation markers (Table 2), paralytic ileus, and low Hct. Therefore, diagnostic imaging control with CT and a digital subtraction angiography (DSA) were performed, showing no signs of major bleeding [Figure 6] and only minor signs of bleeding in the area of the stomach atrium, with concomitant hemorrhagic pancreatitis (Figure 6). The general surgeons performed an investigative laparotomy, intraperitoneal lavage, and finally, a vacuum-assisted closure (VAC) of the abdominal cave was placed. Subsequently, he was transferred to the ICU, where he remained for 20 days. After sepsis was successfully treated, the patient was transferred to the General Surgery Department and he was discharged in good clinical condition after the closure of the abdominal cave without further complications.

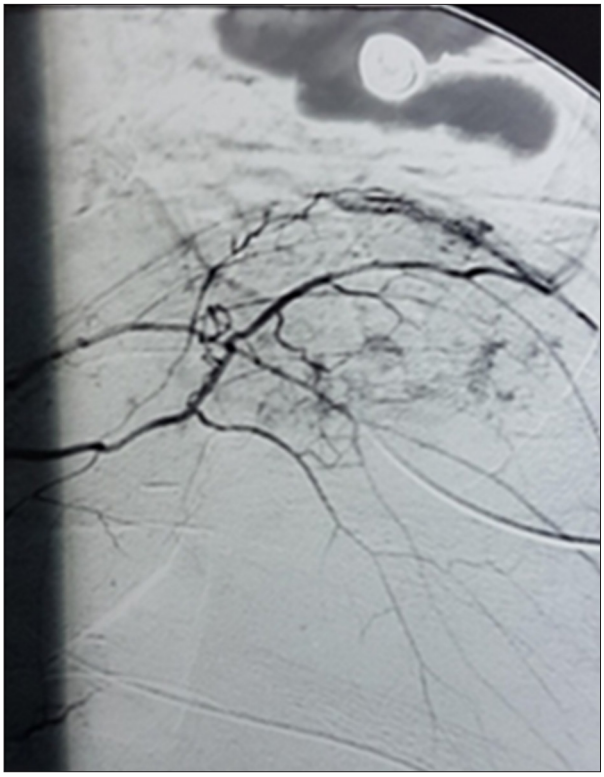

Figure 6. DSA – Minor bleeding in stomach atrium due to pancreatitis.

Discussion

Splenic aneurysms are the most common visceral aneurysms, accounting for 60% of all visceral aneurysms [1]. The overall incidence in the general population is about 1: 10 000. Giant splenic aneurysms are uncommon [3], and there are only 9 reported cases with a concomitant AV-shunt present [4–9]. Hyperdynamic conditions due to arteriovenous shunt lead to portal hypertension and ascites. According to the current literature, acquired splenic arteriovenous shunt is usually iatrogenic or caused by trauma [1] (Table 1). They are 4 times more frequent to women than in men, especially in multiparous women [1]. The vast majority of cases is asymptomatic. Portal hypertension without a concomitant hepatic lesion is present in only 16% of the cases [1]. The clinical presentation of portal hypertension (ascites) derives from congestion of the

Table 2. Inflammatory markers in the 11th and 16th post-op day (hemorrhagic pancreatitis).

Examination	Normal values	Units	Values	
			11th	16th
WBC	3.8–10.5	K/μL	14.26	–
CRP	0.0–0.8	mg/dl	14.200	17.700
PCT	<0.5	ng/ml	–	1.1

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portal circulation, which, if not promptly treated, will lead to an irreversible hepatic lesion if a rupture has not previously occurred. In the case presented here, the giant splenic aneurysm resulted from the commonly ligated long stump of the splenic artery and vein after urgent splenectomy (Figures 1–3). Degenerative erosion of the aneurysm wall due to turbulence and inflammation led to AV-shunt and hyperdynamic portal circulation conditions (Figure 3). Unfortunately, since the case was an emergency, the general surgeons were unable to measure the portal venous pressure. Pancreatitis and paralytic ileus were caused by the giant dimensions of the inflamed omentum sac in contact with the adjacent pancreas. The present case is the 10th to be published [4–9]. Our case is rare and interesting not only due to the extremely high risk of rupture, but also the presence of an A-V shunt leading to portal hypertension, hepatic lesion, and possibly irreversible ascites. Although endovascular treatment remains the best option for repair [2,3], in our case that was excluded due to the high risk of systemic embolization in the portal circulation, which would lead to acute hepatic necrosis and consequent death. Our patient survived and is undergoing rehabilitation in the Department of General Surgery.

Conclusions

Splenic aneurysms are common and require surgical repair (open surgery via aneurysmectomy or endovascular repair via embolization, or a stent graft interposition) [2,3]. Giant splenic aneurysms are rare and are usually caused by a previous splenectomy and preservation of a long vessel stump. These cases need immediate surgical intervention because of high risk of rupture, which would lead to fatal complications.

Department and Institution where work was done

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Conflict of interest

None.

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