



Case Report

Giant Splenic Artery Pseudoaneurysm: A Case Report and Literature Review

Yusuf Yagmur¹, Sami Akbulut², Serdar Gumus¹, Firat Demircan¹

¹*Department of Surgery, Diyarbakir Education and Research Hospital, Diyarbakir, Turkey*

²*Department of Surgery and Liver Transplant Institute, Inonu University Faculty of Medicine, Malatya, Turkey*

Splenic artery aneurysms (SAAs) are the third most frequent intra-abdominal aneurysm, following abdominal aorta and iliac artery aneurysms. SAAs are classified according to their involvement of arterial wall layers: true aneurysms involve all 3 layers (intima, media, and adventitia), and pseudoaneurysms involve only one or two. Herein we present a new case of giant pseudo SAA. A 65-year-old female patient with a pancreatic mass and iron deficiency was referred to our clinic for further investigation. Abdominal ultrasonography, contrast-enhanced CT and magnetic resonance imaging showed a lesion resembling a subcapsular hemangioma in the spleen, and aneurysmatic dilation of the splenic artery with a diameter of >5 cm. The large size of the aneurysm and the clinical findings were indications for surgical treatment. The patient underwent en bloc resection of the spleen, distal pancreas, and aneurysmatic segment of the splenic artery. The patient remains complication-free 2 months after the operation. Spontaneous rupture is the most important life-threatening complications of giant SAAs. Therefore, all symptomatic patients with SAA should be treated, as well as asymptomatic patients with lesions ≥ 2 cm, who are pregnant or fertile, have portal hypertension, or are candidates for liver transplantation. Despite advances in endovascular techniques, conventional abdominal surgery remains the gold standard for treatment.

Key words: Giant splenic artery aneurysm – Open surgery – Splenic artery aneurysms – Chronic pancreatitis

Corresponding author: Sami Akbulut, FICS, FACS, Department of Surgery and Liver Transplant Institute, Inonu University Faculty of Medicine, Malatya 44280, Turkey.

Tel.: +90 422 3410660; Fax: +90 422 3410036; E-mail: akbulutsami@gmail.com

Splenic artery aneurysms (SAAs) account for more than half of all visceral artery aneurysms.¹ Although rare, SAAs can potentially be life threatening, due to spontaneous intraperitoneal rupture, rupture into the neighboring hollow organs, and fistulization into the pancreatic duct.^{1,2} Most small SAAs are asymptomatic, and are therefore diagnosed incidentally during radiologic investigations.^{3–6} In contrast, giant (≥ 5 cm) SAAs are symptomatic and can result in complications, and may be detected as a pulsatile mass in the upper-left quadrant or epigastrium. The most frequent symptoms for symptomatic cases are epigastric and left-upper quadrant abdominal pain, but patients can also present with general gastrointestinal complaints of a feeling of fullness, loss of appetite, nausea, or vomiting.^{4,7} Herein we report a new case of giant SAA, which is among the largest reported in the literature.

Case Report

A 65-year-old female patient was referred to our surgery clinic because of abdominal pain, dyspepsia, fatigue of 3 months' duration, iron deficiency, and a pancreatic mass. Abdominal ultrasonography (US) showed a 101×55 mm cystic lesion in the pancreatic tail. Intravenous contrast-enhanced CT showed a lesion resembling a subcapsular hemangioma in the spleen, and aneurysmatic dilation of the splenic artery with a maximum dimension of 10×5 cm (Fig. 1). T2A-weighted magnetic resonance imaging (MRI) detected an aneurysmatic dilation with a diameter >5 cm, and a hyperintense hemangioma in the spleen (Fig. 2). The clinical

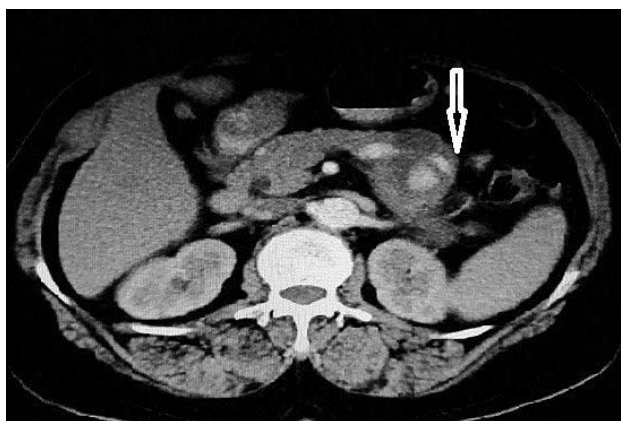


Fig. 1 Contrast-enhanced CT showing an aneurysmatic dilation (10×5 cm) of the splenic artery.



Fig. 2 T2A-weighted magnetic resonance imaging revealed an aneurysmatic dilation >5 cm arising from the splenic artery.

findings and dimensions of the aneurysm were indicators for surgical treatment. The operation was initiated via a midline abdominal incision for a laparotomy. A pulsatile, 10×6 cm splenic artery aneurysm was located on the pancreatic tail, filling the bursa omentalis cavity, with dense adhesions to the adjacent pancreatic tissue. Multiple peripancreatic and perisplenic venous dilations, primarily on the right gastroepiploic vein, were inspected (Fig. 3). Following incision of the gastrocolic ligament, the aneurysmatic portion of the splenic artery was observed, revealing a larger dimension than was seen with CT. The splenic artery was tortuous, with dense adhesions between the artery and distal pancreas. The findings were concordant with chronic pancreatitis, thus explaining the pathogenesis of the adhesions. Consequently, en bloc resection of the



Fig. 3 Multiple peripancreatic and perisplenic venous dilations, primarily on the right gastroepiploic vein, were inspected.

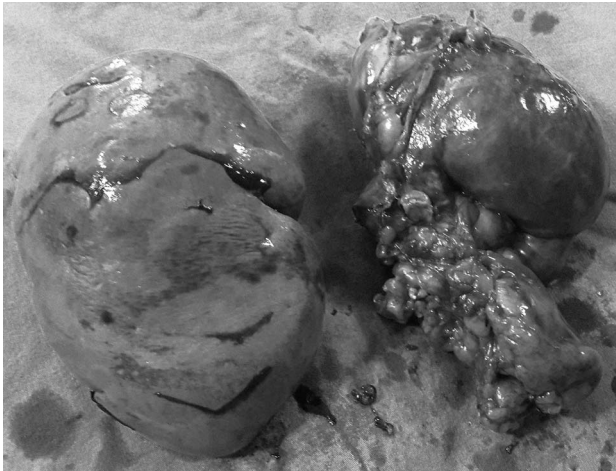


Fig. 4 En bloc resection of the spleen, distal pancreas, and aneurysmatic segment of the splenic artery was performed.

spleen, distal pancreas, and aneurysmatic segment of the splenic artery were performed (Fig. 4). The postoperative course was uneventful, and the patient was complication-free 2 months after the procedure.

Discussion

SAAs constitute 50 to 70% of splanchnic artery aneurysms,^{8–10} and are the third most frequent intra-abdominal aneurysm, after aneurysms of the abdominal aorta and iliac artery.^{4,11} The incidence of SAA is unclear, and various studies of autopsies and angiography indicate rates of 0.01 to 0.20% and 0.78 to 0.80%, respectively.^{1,5,8,11–14} However, SAAs are 4 times more frequent in women, and are most commonly diagnosed in individuals older than 50 years of age.^{1,8,9}

The pathogenesis of SAA is not well understood, but involves the loss of the media layer, characterized by disintegration of elastic fibers and loss of smooth muscle.^{1,8,15} SAAs are classified by histopathology as either true aneurysms (vascular enlargements involving the intima, media, and adventitia layers of the artery wall) or pseudoaneurysms (enlargements that do not involve all three artery wall layers).¹⁵ SAAs typically measure 2.1 cm in diameter at the time of diagnosis, and rarely exceed 3 cm^{5,8,16}; those measuring >5 cm are generally described as giant.^{1,6,16–19}

Risk factors for true SAAs are hypertension, atherosclerosis, cirrhosis, portal hypertension, liver transplantation, female sex, pregnancy, and multiparity,^{4,6,8,14,20} but can also include splenomegaly,

medial fibrodysplasia, arteritis, collagen vascular disease, polyarteritis nodosa, systemic lupus erythematosus, anomalous splenic artery origin, α 1-antitrypsin deficiency, and inflammatory and infectious diseases.^{7,20} In contrast, pancreatitis (chronic or acute) is the primary risk factor for pseudo-SAAs, along with pancreatic pseudocysts and abdominal trauma.^{4,19,21} Pseudoaneurysms can develop as a result of the disintegration of elastin fibers by pancreatic enzymes following arterial wall destruction.^{15,18,21} Indeed, the patient in the present case suffered from chronic pancreatitis.

Diagnostic tools for SAAs include US, Doppler US, CT, and MRI.^{5,16} Doppler US is the first choice for pregnant patients,⁴ and CT and CT angiography are useful for observing the typical aneurysm body in the arterial phase.^{6,15} Although CT requires radiation exposure and involves risk of contrast-nephropathy, it is beneficial in differentiating SAAs from pancreatic tumors, pseudocysts, solid epithelial tumors, and gastric leiomyomas. On the other hand, MRI is more sensitive and specific, but is contraindicated for patients with pacemakers, metal prostheses, can induce emotional and respiratory problems for claustrophobic and respiratory-distressed patients, particularly with respect to the prolonged time requirement, and the availability is often limited to an emergency basis.⁴

Although there is no consensus on the management of SAA patients, there have been major changes as a result of progress in radiologic diagnosis and treatment options.¹ Irrespective of the diameter, treatment is recommended for all symptomatic SAAs, as well as for asymptomatic patients with lesions ≥ 2 cm (due to high risk of rupture), who are pregnant or fertile, have portal hypertension, or are candidates for liver transplantation.^{1,3,4,7,8,10} In comparison, the rupture risk for pseudo-aneurysms is not related to their dimensions, and all pseudoaneurysms should be treated.¹⁷

Management of SAAs depends on their dimensions, location, complications, and severity of the clinical findings,^{1,5,15,18} and most frequently involves open abdominal surgery, endovascular treatment (coil embolization or stent), laparoscopic surgery, and medical treatment.^{1,5,7–9,17,22,23} Proximally located, elongated, and tortuous SAAs are suitable for aneurysmectomy and end-to-end reconstruction without splenectomy,⁴ though splenectomy might be added for lesions originating from the distal two-thirds of the splenic artery.^{9,15} For giant SAAs or cases where a simple aneurysmectomy is impossible due to dense strictures, preferred treat-

ment options include an aneurysmectomy plus splenectomy, bipolar splenic artery ligation with or without aneurysmectomy, transaneurysmal splenic artery ligation, or distal pancreatectomy when necessary.^{4,5} The patient in the case presented here underwent an aneurysmectomy plus splenectomy along with a pancreatectomy because dense adhesions had developed between the aneurysm and distal pancreas.

Laparoscopic SAA excision is a minimally invasive alternative to open abdominal surgery,⁵ particularly for use during early pregnancy and with small lesions.⁸ However, it is contraindicated in hemodynamically unstable patients or those at rupture risk, and is not suitable for larger aneurysms and lesions with dense adhesions to surrounding tissues.

Endovascular treatment options, such as transcatheter embolization, percutaneous injection, and endovascular stent grafts,¹⁰ are increasingly favored due to their acceptable technical success and low morbidity rates.^{4,9,17} Embolization is the first option for asymptomatic lesions diagnosed incidentally, whereas transcatheter embolization is preferred in cases involving surgical technical difficulty and in patients at increased operative risk. The most frequent complications of transcatheter embolization are coil migration, aneurysm rupture, intestinal infarct, fever, splenic infarct, and abscess formation.^{12,17,18} If embolization fails or is unsuitable, percutaneous injection may be used, which involves direct coil application or thrombin injection into the lesion.^{10,24} Most recently, endovascular stenting has been introduced as a treatment for SAAs.⁵ This minimally invasive technique preserves splenic perfusion by placing a stent (self-expanding and balloon-expanding) in the aneurysm without dilating the aneurysmatic segment, and minimizes splenic infarction and abscess complications that occur with coil embolization.¹⁰ However, as with embolization, the application of this technique is limited by tortuous arteries, decreased artery dimensions, and the location of the lesion.¹⁷

There is a limited number of studies concerning optimum treatment option for SAA. The last updated study was made by Hogendoorn *et al.*²⁵ Its authors revealed the superiority of endovascular treatment to open surgery in terms of perioperative mortality and short-term outcomes. However, authors stated that open surgery has advantages for both long-term outcomes and re-intervention options during follow-up.²⁵

Spontaneous rupture and fistulization into neighboring organs are the most important life-threatening complications of giant SAAs. Therefore, all complicated or symptomatic patients with SAA should be treated, as well as asymptomatic patients with lesions ≥ 2 cm, who are pregnant or fertile, have portal hypertension, or are candidates for liver transplantation. Despite advances in endovascular techniques, conventional abdominal surgery remains the gold standard for treatment.

Acknowledgments

The authors declare that they have no conflict of interest.

References

1. Uyar IS, Okur FF, Akpınar BA, Abacılar F, Yurtman V, Sahin V. Giant splenic artery aneurysm: a case report [in Turkish]. *Türk Göğüs Kalp Damar Cerrahisi Dergisi* 2013;**21**(3):799–802
2. Orsitto G, Fulvio F, Pinto AG, Turi V, Tria D, Venezia A. Geriatric assessment of a giant splenic artery aneurysm accidentally diagnosed. *Aging Clin Exp Res* 2011;**23**(5-6):491–494
3. Rathod J, Taori K, Dhokane S, Pawar P, Vaghasiya D. Endovascular embolisation of giant ruptured proximal splenic arterial pseudoaneurysm using microcoils & glue: case report. *Surg Sci* 2011;**2**:290–293
4. Mishra PK, Saluja SS, Sharma AK, Pattnaik P. Management of splenic artery aneurysm associated with extrahepatic portal vein obstruction. *Hepatobiliary Pancreat Dis Int* 2012;**11**(3):330–333
5. Yadav S, Sharma P, Singh PK, Punia S, Desai P, Anjan AK. Giant splenic artery aneurysm: a rare but potentially catastrophic surgical challenge. *Int J Surg Case Rep* 2012;**3**(11):533–536
6. Goldberg RF, Maley W, Kennedy EP. Giant splenic artery pseudoaneurysm. *J Gastrointest Surg* 2011;**15**(6):1063–1066
7. Pappy R, Sech C, Hennebry TA. Giant splenic artery aneurysm: managed in the cardiovascular catheterization laboratory using the modified neck remodeling technique. *Catheter Cardiovasc Interv* 2010;**76**(4):590–594
8. Bakhos CT, McIntosh BC, Nukta FA, Fiedler PN, Denatale RW, Sweeney TF. Staged arterial embolization and surgical resection of a giant splenic artery aneurysm. *Ann Vasc Surg* 2007;**21**(2):208–210
9. Sachdev-Ost U. Visceral artery aneurysms: review of current management options. *Mt Sinai J Med* 2010;**77**(3):296–303
10. Goes AM Jr, Góes AS, de Albuquerque PC, Palácios RM, Abib Sde C. Endovascular treatment of giant splenic artery aneurysm. *Case Rep Surg* 2012;**2012**:964093

11. Aroor AR, Prakasha SR, U R, Attar NR. Multiple splenic artery aneurysms: a rare cause of extrahepatic portal hypertension and massive splenomegaly. *J Clin Diagn Res* 2014;**8**(9):MD01–MD02
12. Miao YD, Ye B. Intragastic rupture of splenic artery aneurysms: three case reports and literature review. *Pak J Med Sci* 2013;**29**(2):656–659
13. Law ST, Wong CK, Chow KC, Loo KT. Splenic arteriovenous fistula: unusual cause of portal hypertension complicated with gastric variceal bleeding. *J Dig Dis* 2012;**13**(10):549–552
14. Mastroroberto M, Berardi S, Renzulli M, Maggoli C, Pianta P, Pinna AD. Transcatheter embolization for giant splenic artery aneurysms: still an open question. *Case Rep Radiol* 2012;**2012**:652469
15. Ho MF, Chan YC, Cheng SW. Successful endovascular management of giant splenic artery aneurysms. *Vascular* (in press)
16. Manian UD, Badri H, Coyne PE, Nice CA, Ashour HY, Bhattacharya V. Endovascular treatment of a ruptured splenic artery aneurysm using Amplatzer vascular plug. *Int J Biomed Sci* 2009;**5**(1):81–84
17. Al-Habbal Y, Christophi C, Muralidharan V. Aneurysms of the splenic artery - a review. *Surgeon* 2010;**8**(4):223–231
18. Agrawal GA, Johnson PT, Fishman EK. Splenic artery aneurysms and pseudoaneurysms: clinical distinctions and CT appearances. *AJR Am J Roentgenol.* 2007;**188**(4):992–999
19. Karsidag T, Soybir G, Tuzun S, Makine C. Splenic artery aneurysm rupture. *Chirurgia (Bucur)* 2009;**104**(4):487–490
20. Tirpude B, Bhanarkar H, Dakhore S, Surgule D. Giant splenic artery pseudo aneurysm masquerading as bleeding per rectum - a rare case. *J Evol Med Dent Sci* 2013;**2**(44):8569–8573
21. Tekola BD, Arner DM, Behm BW. Coil migration after transarterial coil embolization of a splenic artery pseudoaneurysm. *Case Rep Gastroenterol* 2013;**7**(3):487–491
22. Gupta V, Kumar S, Kumar P, Chandra A. Giant pseudoaneurysm of the splenic artery. *JOP* 2011;**12**(2):190–193
23. Ali S, Verma V, Sastry R, Wani I. Giant splenic artery aneurysm: case report. *ISRN Surg* 2011;**2011**:383450
24. Huang IH, Zuckerman DA, Matthews JB. Occlusion of a giant splenic artery pseudoaneurysm with percutaneous thrombin-collagen injection. *J Vasc Surg* 2004;**40**(3):574–577
25. Hogendoorn W, Lavida A, Hunink MG, Moll FL, Geroulakos G, Muhs BE. Open repair, endovascular repair, and conservative management of true splenic artery aneurysms. *J Vasc Surg* 2014;**60**(6):1667–1676