



Case Report

Splenic artery aneurysm masked as a gastroenterology complication: A case report and literature review

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ABSTRACT

Introduction and importance: Splenic artery aneurysm has an insidious onset, and low incidence, most of which have no specific manifestations on the early onset and remains the most common visceral aneurysm and third most common splanchnic aneurysm as it still remains a challenge to deal with clinically by many clinicians.

Case presentation: We report a single case of a young 21 years old girl who had no potential risk of splenic artery aneurysm on clinical presentation, for gastroenterology disease only assessment and attention in our facility. The patient born and raised on a tropical island in Southern China was clinically diagnosed with splenic artery aneurysm-associated gastroenterological complications which was presented earlier as hematemesis. The patient was considered to have received optimal critical care by our multidisciplinary team and classical features displayed within the clinical settings are worth documenting and contribute perfectly to medical literature as the patient on follow-up is now back to normal life.

Clinical discussion: Our patient recovered excellently on critically close follow-up since the patient had special gastroenterology associated complication features which masked the splenic artery aneurysm with very encouraging post-operative parameters or results.

Conclusion: The patient was considered to have received optimal multidisciplinary quaternary medical care for SAAs with gastroenterology-associated complications in our interventional cardiovascular and gastroenterology medicine department.

1. Introduction

Splenic artery aneurysm(SAA) is not relatively rare in clinical practice, but it has the potential risk of massive abdominal hemorrhage when rupture ensues on the onset. Clinically, splenic artery aneurysm(SAA) is defined as a condition where there is a focal dilation in the diameter of the splenic artery that is 50% greater than the normal vessel diameter. From histopathology, it can be divided into two types: true aneurysms and false aneurysms (pseudoaneurysms). The size and number of splenic arteries vary greatly among individuals, but more than 60% of patients with splenic artery aneurysms are isolated and less than 2 cm in diameter, 75% are located in the distal 1/3 of the splenic artery, and about 20% are located in the middle 1/3 of the splenic artery [1]. The diameter

of splenic aneurysms rarely exceeds 3cm, and most patients with splenic aneurysms have no obvious clinical manifestations [2]. They are always accidentally discovered during abdominal CT or B-ultrasound examinations through monitoring of doppler signals, although with CT and digital subtraction angiography (DSA) examination techniques, the discovery rate has gradually increased. Those with a diameter of more than 5 cm are called giant splenic aneurysms, and giant splenic aneurysms can have clinical symptoms such as abdominal distention and abdominal pain, and there is a greater risk of rupture. Once the splenic aneurysm ruptures and hemorrhages, intra-abdominal hemorrhage or gastrointestinal hemorrhage may occur, and the mortality rate is as high as 25%–70% [3]. Therefore, early diagnosis and effective interventions are very important for the good prognosis of patients. The concept of

Abbreviations: SAA, splenic artery aneurysm; DSA, digital subtraction angiography; BP, blood pressure; CTA, computed tomography angiography.

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splenic artery aneurysm(SAA) has been well established and documented in medical literature. The most common causes of tumor rupture are acute pancreatitis, chronic pancreatitis, atherosclerosis, hypertension, liver cirrhosis, trauma, and pregnancy, among which pregnancy is the most important factor [2–4]. Patients with an obvious risk of rupture should be treated immediately with surgical or interventional care. It is generally believed that those with an aneurysm diameter greater than 2 cm have a greater risk of rupture, and active treatment is recommended. For pseudo-aneurysms, the risk of rupture has nothing to do with the size of the inner diameter, and all pseudo-aneurysms should be actively treated [5]. CT remains the most preferred golden standard imaging method for preliminary diagnosis of splenic artery aneurysms although other advanced imaging gives clinicians and surgeons an upper hand on perfect surgical or therapeutic approach to resolving or excluding the clinical condition. Relevant literature reports that CTA technology can directly be used to diagnose visceral aneurysms (including SAA), and that its accuracy is also very high [6–12]. We primarily focused our attention on presenting a single patient experience of splenic artery aneurysm which was masked as primary gastroenterology associated complication which occurred in a patient with relatively ‘low’ to ‘no’ risk of developing splenic artery aneurysm who was given excellent care in an advanced quaternary medical care center. Knowledge of the wide range of masked clinical conditions of splenic artery aneurysms and treatment protocol is primarily essential in effective and timely treatment or exclusion of the clinical condition.

1.1. Clinical case presentation

Our work consists of a single case report and has been reported in accordance with SCARE 2020 criteria [13].

A 21-year-old female patient presented to our hospital on November 08, 2018, due to “intermittent upper abdominal pain with hematemesis for 3 weeks”. The patient had a past 3 weeks before presentation to our hospital with abdominal pain which was obvious after meals, lasted longer, no radiating pain, accompanied by hematemesis, complained of blood clots of unknown amount when she visited the washroom which is classical of melena, no abdominal distention, dizziness, and fatigue. The patient had no drug history and no significant family-associated genetic diseases.

On general physical examination, She presented with signs of hypovolemic shock with paleness, sweating, low blood pressure (BP of 108/69 mmHg, P 82 beats/min, R 20 beats/min, T36.6 °C), and acute anemia (hemoglobin level of 6.9 d/dL on the first check). The patient had a clear mental state, flat and soft abdomen, negative mobile dullness, and bowel sounds of 4 beats/min were noted.

Electronic gastroscopy on the 8th of November 2018 performed by our team showed chronic non-atrophic gastritis, and biliary hemorrhage. Routine blood work up showed red blood cells, $2.31 \times 10^{12}/L$, hemoglobin, 69g/L, hematocrit, 0.210L/L, platelet, $61 \times 10^9/L$; D-dimer 1.87mg/L, with a preoperative enhanced CT of the abdomen which suggested splenic pseudoaneurysm and chronic pancreatitis Figs. 1 and

2.

An esophagogastroduodenoscopy (EGD) was performed in the gastroenterology emergency setting by the team (composed of specialists, postgraduate residents, general practitioners and others), with a standard gastroscope (Olympus Medical System® GIF-Q160).

Endoscopy showed the presence of a pulsatile bulging at the proximal third of the greater curvature posterior wall of the corpus of the stomach (Fig. 5c). The gastric bulging measured about 3 cm in diameter and presented a small erosion on its surface with a visible vessel with slow active bleeding at the point of the examination, despite the gastric cavity being quite full of fresh blood. An endoscopic cyanoacrylate glue injection was performed (n-Butyl 2-cyanoacrylate, Glubran®2, GEM, Italy): The glue was delivered into the lesion through a 23-gauge injection needle catheter (Cook-Medical Inc.), followed by injection of 1.2 ccs of sterile water. No other lesions of the upper GI tract were detected during the EGD.

Esophagogastroduodenoscopy findings. Fig. 6. A: Bulging at the proximal third of greater curvature/posterior wall of the stomach (arrow); B and C: Minute erosion on the bulging surface with a visible vessel (arrow); D: endoscopic cyanoacrylate glue injection into the lesion through a 23-gauge needle catheter (+).

The clinical scenario and the finding presented led us to perform an urgent contrast-enhanced computed tomography (CT) after the EGD. CT angiography (Fig. 2), disclosed that the middle and distal third of the splenic artery was fully replaced by an aneurysm (9.85 mm in maximum diameter); it was packed to the posterior wall of the stomach, with a plugged fistula. No active bleeding was noted, confirming that EGD treatment was successful. The opacification of the aneurysm was partial, because of its large volume and low pressure. Contrast-enhanced computed tomography angiography showed a large mass, with the partially calcified wall, adjacent to the spleen; Fig. 2 a and b: The opacification of the aneurysm is partial and low, because of its large volume and low pressure. It is packed to the posterior wall of the stomach (arrow head); Fig. 2b. After contrast injection, CT showed a splenic artery fully replaced by an aneurysm (32.88mm in maximum diameter, panel, coronal view), partially thrombosed. Fig. 2b.

Electronic gastroscopy on the 8th of November 2018 showed chronic non-atrophic gastritis and biliary hemorrhage. Fig. 6. The CT angiography(CTA) (Fig. 2) also confirmed the middle and distal third of the splenic artery were fully replaced by a splenic artery aneurysm (SAA) (32.88mm in maximum diameter) with a closely normal spleen and normal pancreas. Aneurysmal dilatation of the middle-distal part of the splenic artery was measuring 32.88mm in diameter and it could be considered true since it was fusiform, involving all the wall layers and with no surrounding inflammation.

The patient received hemagglutinin, perioperative anti-microbial therapy, red blood cell suspension, endoscopy guided upper gut repair and hemostasis therapy (FUJINON NKJ 180018869), splenic artery embolization, and trunk embolization therapy in our interventional gastroenterology and cardiovascular medicine department on November 19, 2018, with additional fluid rehydration therapy and other

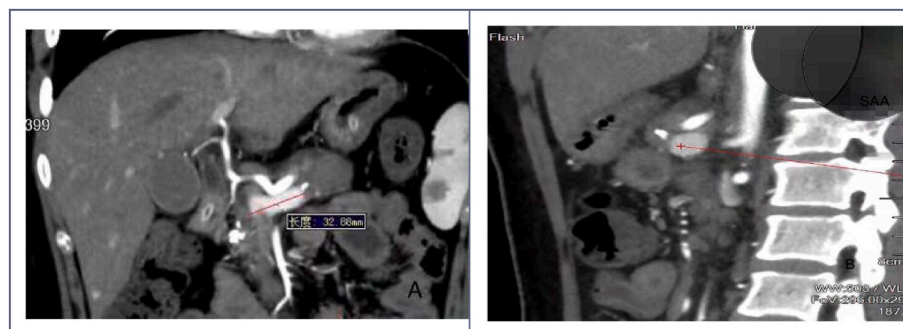


Fig. 1. Preoperative abdominopelvic CT Imaging of splenic artery aneurysm (SAA).

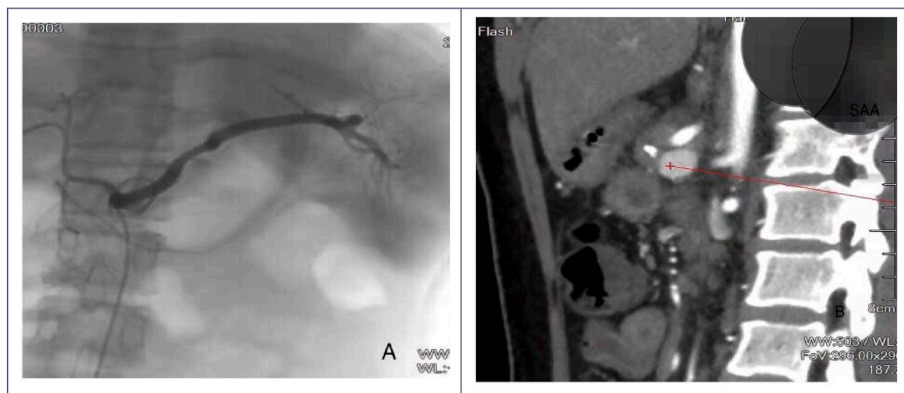


Fig. 2. Preoperative CT angiography imaging.

symptomatic treatment. The procedure was executed by the team (composed of specialists, postgraduate residents, general practitioners and others) via the modified seldinger technique and 5F femoral artery sheath (Cordis, Tipperary, Ireland) under the guidance of a 0.035" super-smooth guidewire. Angiography with a Yashiro catheter showed that the spleen was normal in size and shape, the wall of the proximal splenic artery was irregular, nodular pseudoaneurysm was seen in the dorsal pancreatic artery area, and the parenchyma of the spleen was well developed. Figs. 4 and 5. With the help of the 2.6F micro-catheter (Progreat®, Terumo, Leuven, Belgium), eight(8) micro coils (10mm × 14mm) 3 pieces each, (8mm × 12mm) 3 pieces each, (6mm × 6.5mm) 12 pieces each (Interlock-18, Boston Scientific, Cork, Ireland), and N-butyl-cyanoacrylate (NBCA) glue (Histoacryl®, Brown, Hessen, Germany) were inserted through the micro-catheter. The main splenic artery embolization was performed on the line segment (about 2mm) Fig. 2. The second angiography showed that the main splenic artery was successfully embolized and the blood flow in the remaining lumen was thin and tortuous Fig. 5. It was then keenly monitored primarily focusing on the vital signs perioperatively; as immobilization of the right lower limbs for 12 hours was performed, compression of the puncture site for 6 hours was done simultaneously as continuous infusions of acid suppression and hemostasis after the operations were performed. The patient also received postoperative symptomatic, prophylactic antimicrobial therapy and supportive care. The postoperative condition is currently relatively stable, and the hemoglobin fluctuates between 89–100g/L. As of 2018/11/23 upper abdominal plain scan and enhanced MRI showed the splenic aneurysm was significantly smaller than before (Reducing significantly from 32.88mm in maximum diameter to 9.85mm as shown in Figs. 3–5) She was discharged from the hospital 10 days after the operation.

1.2. Outcome and follow-up

The length of postoperative care was two weeks, without complications. During the hospital stay, the patient underwent radiological and clinical work-up. Total body multi-slice CT scan with IV contrast was performed to exclude any other site of visceral aneurysms (chest and brain). Diseases such as arterial and portal hypertension, atherosclerosis, diabetes, alpha-1-antitrypsin deficiency, liver cirrhosis, and collagenopathy were ruled out.

2. Discussion

Several mechanisms have been proposed to have a characteristic contributive role in the pathogenesis of splenic artery aneurysms. Proposals made by Trimble and Hill [8–12] remain valid, since they suggest that an aneurysmal dilatation is the result of two contributing factors: weakness of the artery wall and increased blood pressure. The most often reported risk factors are: portal hypertension; arterial hypertension [11,12]; atherosclerosis; and diabetes [8–13]. The exact etiology of visceral aneurysms is not established. Recent literature has suggested that true aneurysms develop secondary to arterial wall weakness due to several causes. These include atherosclerosis (32%), medial degeneration or dysplasia (24%), abdominal trauma (10%), hypertension, connective tissue diseases, and necrotizing vasculitides such as polyarteritis nodosa or Wegner granulomatosis Table 1 [14–23]. Endovascular and endoscopic minimally invasive surgical treatment is a safe, effective, minimally invasive therapeutic option for splenic artery aneurysms (SAAs) and its associated gastric abnormality or complications, as these reliable therapeutic options allow patients considered medically unfit for traditional surgical open repair to be treated. The choice of the most suitable materials depends on the anatomical characteristics of these aneurysms. However, further explorative and innovative studies are

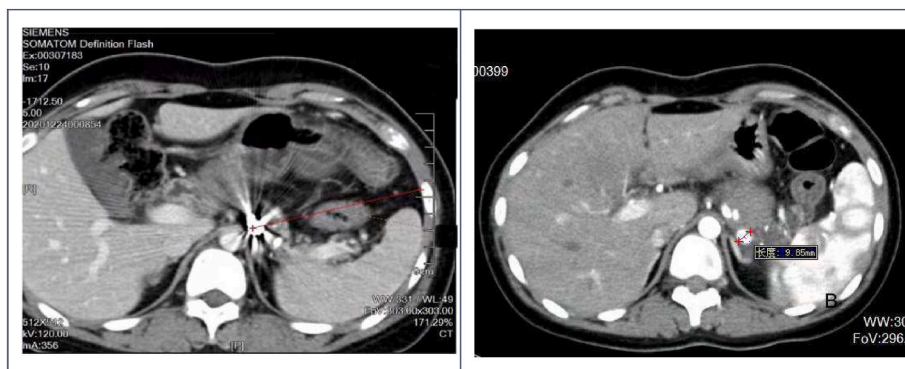


Fig. 3. Postoperative CT imaging.

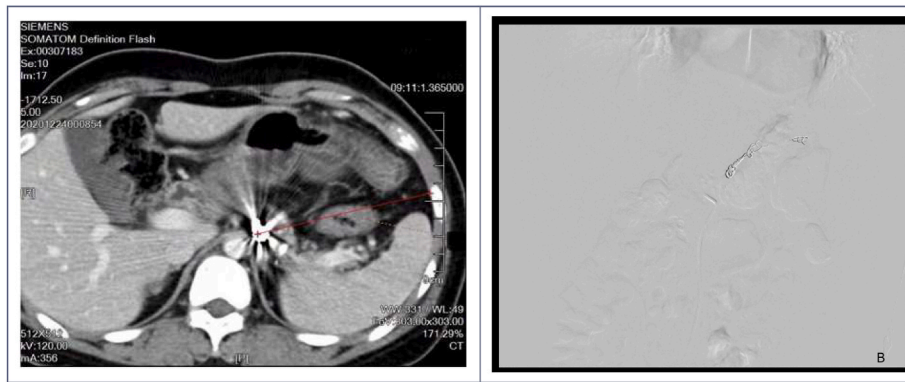


Fig. 4. Postoperative CT imaging.

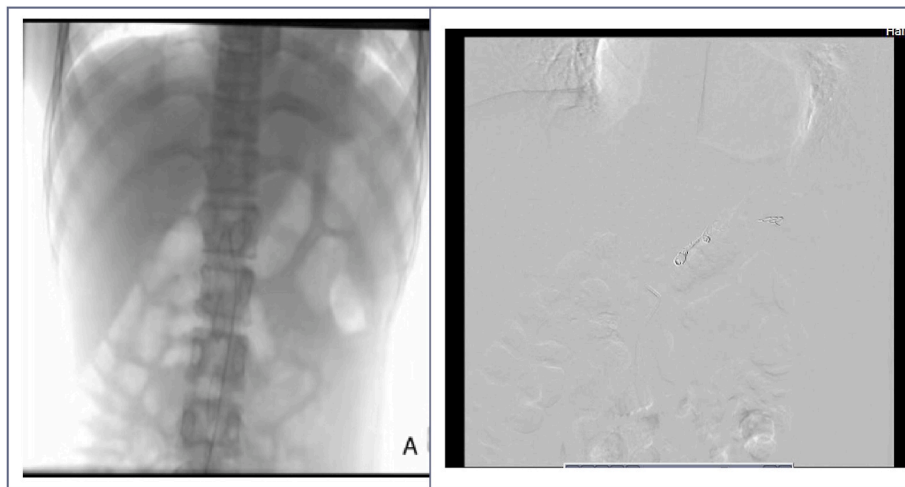


Fig. 5. Postoperative CT imaging.

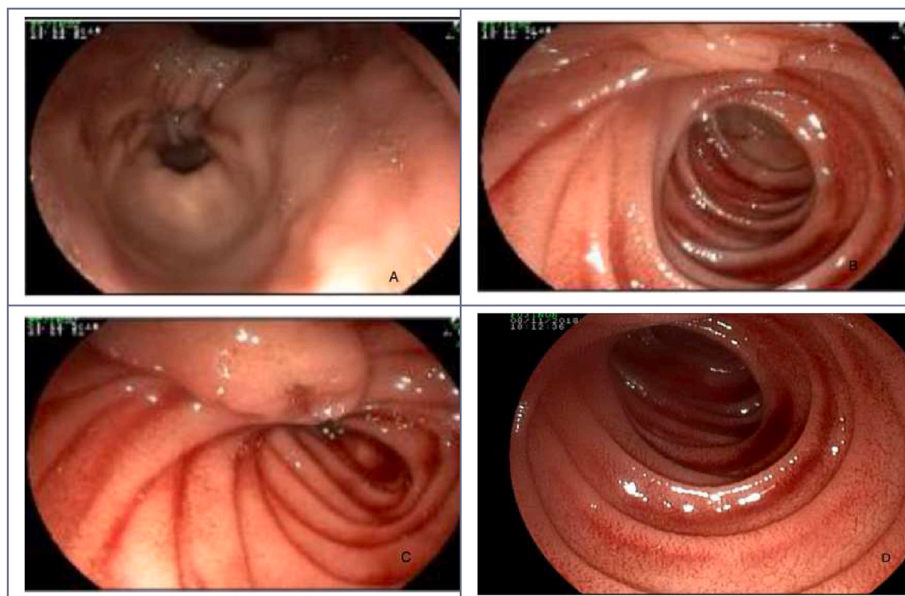


Fig. 6. Gastroscopy plus hemostasis therapeutic procedure.

needed to determine the long-term clinical efficacy and applicability of these therapeutic techniques.

We believe that surgical treatment of aneurysms should also be

actively performed to reduce the mortality of patients. The key to surgical treatment of splenic artery aneurysms is to totally exclude the SAAs. At present, there are two main ways to treat splenic aneurysms:

Table 1
Summary of general patient characteristics of some previously reported cases of splenic artery aneurysm(SAA).

Case Report/ Series	Name of Author	No. Of Cases	Year of Publication	Sex(M/ F)	Age	Rupture/ NonRupture	State	Time interval between initial presentation & Rupture of SAA	
Case Report	Ceccarelli G et al.	1	2020	F	65	No	Not Pregnant	N/A	
Case Report	Fujii M et al.	1	2020	F	40	Yes	Pregnant	6 hours	
Case Report	Panzer F et al.	1	2020	M	35	No	Not pregnant	N/A	
Case Report	Colsa-Gutiérrez P	1	2015	F	40	No	Not Pregnant	3 Weeks	
Case Report	Khan A et al.	1	2017	M	50	Yes	Not Pregnant	24 hours	
Case Report	Guang LJ	1	2015	F	54	No	Not Pregnant	N/A(Alive)	
Case Report	De Silva WSL	1	2017	M	60	Yes	Not Pregnant	24 hours(Alive)	
Case Report	Chen G	1	2019	M	50	Yes	Not Pregnant	4 hours	
Case Series	Oakley E	2	2013	M	26.5	Yes(all)	Splenectomy and aneurysms resection	Not Pregnant	N/ A

one is interventional therapy, and the other is surgical treatment. Other treatment methods are based on the above-mentioned methods.

The surgical procedure of a splenic artery aneurysm depends on the site of the aneurysm.

- ① When the SAA is far away from the hilum of the spleen, at the beginning of the splenic artery. This type of splenic aneurysm can preserve the spleen, that is, the proximal and distal segments of the aneurysm can be resected utilizing arterial ligation or resection of aneurysm and splenic artery reconstruction [8];
- ② If the SAA is tightly attached to the body and tail of the pancreas, it is difficult to ligate the artery, and forced separation is likely to cause hemorrhage, so combined spleen and pancreatic body and tail resection is feasible;
- ③ If the SAA is close to the splenic hilum, only spleen and splenic aneurysm resection can be performed;
- ④ If there is an internal fistula between the splenic aneurysm and the portal vein, the blood supply of the aneurysm should be blocked first, then the fistula should be repaired, and so the SAA can be removed. Portal hypertension is accompanied by splenic aneurysm. In addition to treating the aneurysm, it is necessary to treat the complications of portal hypertension, such as portal azygos de-vascularization, splenorenal shunt, and more importantly gastroenterology complications.

The celiac artery angiography in our case scenario showed that the splenic artery was locally expanded like a tumor. Figs. 1 and 2. The interventional embolization therapy of the splenic artery is widely used in the treatment of splenic aneurysms and has achieved excellent promising results. The reasons for some complications (such as liver abscess, bleeding at the puncture site), and easy recurrence should be determined after a comprehensive evaluation. An aneurysm was visible in the main trunk, local contrast agent was retained, and then multiple coils were placed to embolize the distal trunk of the splenic artery Figs. 4 and 5, along the aneurysm breach and the proximal trunk of the splenic artery. This case which we handled successfully although challenging on presentation clinically had a relatively fair postoperative recovery without special complications. In summary, CT examinations and confirmatory monitoring of doppler signals via abdominal ultrasonography should be routinely performed for patients with gastroenterology presentations, classically hematemesis associated splenic artery aneurysm complications. Routine clinical evaluation is also very important for improving the survival rate of patients. We proposed a hypothesis that the diagnosis of SAA should be excluded in cases of recurrent upper gastrointestinal bleeding with no other pathological evidence or when a suspected gastric lesion is detected after massive hematemesis in an otherwise healthy patient.

3. Conclusion

Splenic artery aneurysm (SAA) although rare in the general population is gradually becoming common in the general population in recent times. True SAAs can result in intragastric rupture with catastrophic GI bleeding. Splenic artery aneurysms can result in recurrent upper gastrointestinal bleeding. The possibility of impending catastrophic bleeding should be remembered when managing patients with splenic artery aneurysms after a minor bleeding. Negative endoscopy and ultrasonography should require contrast-enhanced computed tomography angiography to look for the cause of recurrent upper gastrointestinal bleeding and a follow up confirmatory doppler ultrasonography. Splenic artery aneurysms (SAAs) can be masked by chronic non-atrophic gastritis, and biliary hemorrhage. Advancement in the field of medicine within a clinically advanced multidisciplinary setting enables an extremely effective and reliable platform in addressing gastroenterology associated complications in splenic artery aneurysm patients.

Ethical approval

The study methods described were performed in accordance with CARE guidelines and other relevant regulations. The study was conducted at the Department of Gastroenterology, The Second Affiliated Hospital of Hainan Medical University. Written informed consents were obtained from clinical research patient or subject before enrolment on case study. Approval was granted by the Ethics Committee and Institutional Review Board of The Second Affiliated Hospital of Hainan Medical University. The procedures used in this study adhere to the tenets of the Declaration of Helsinki.

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Author contribution

Author Contributions: ZJL,YW, JL, JWA, LW, and FBH: Conceptualization, formal analysis, funding acquisition, software, and writing – review and editing. ZJL,YW, JL, JWA, LW, and FBH, and JWA.: Data

curation, methodology, project administration, writing – original draft, and writing – review and editing.

Research registration number

1. Name of the registry: Research Registry.
2. Unique Identifying number or registration ID: research registry7449.
3. Hyperlink to your specific registration (must be publicly accessible and will be checked): <https://researchregistry.knack.com/research-registry#home/registrationdetails/61b616309566d4001e2b39c1/>.

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Consent

Written informed consent was obtained from the patient for publication of this Case report and any accompanying images. A copy of the written consent is available for review by the Editor of this journal.

We also do provide assurance that alterations.

Provenance and peer review

Not commissioned, externally peer reviewed.

Declaration of competing interest

There are no conflicts of interest.

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