Am J Transl Res 2021;13(7):8172-8178
www.ajtr.org /ISSN:1943-8141/AJTR0128295

Original Article

Superior mesenteric arteriovenous fistula presenting as diarrhea: a case report and literature review

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Received December 15, 2020; Accepted March 25, 2021; Epub July 15, 2021; Published July 30, 2021

Abstract: Background: Superior mesenteric arteriovenous fistula (SMAVF) is a very rare disease and mainly manifests as abdominal pain, diarrhea, anorexia, and other portal hypertension symptoms. The diagnosis of the disease mainly relies on abdominal enhanced CT+3D reconstruction or digital subtraction angiography, and the treatment is mainly vascular interventional fistula occlusion. Case summary: a 17-year-old female with a history of abdominal trauma and surgery was admitted to our hospital for diarrhea and abdominal distension. The patient was diagnosed with a superior mesenteric arteriovenous fistula after abdominal enhanced CT + 3D reconstruction. The patient was satisfied with the results after the superior mesenteric artery angiography + covered stent implantation. No discomfort symptoms occurred during the two-year follow-up. Conclusion: A history of abdominal trauma or surgery and clinical manifestations in combination with a radiological analysis are important indicators in the diagnosis of SMAVF. Interventional therapy is the preferred treatment.

Keywords: Superior mesenteric arteriovenous fistula (SMAVF), diarrhea, portal hypertension

Introduction

Superior mesenteric arteriovenous fistula (SMAVF) is a very rare disease and mainly manifests as abdominal pain, diarrhea, anorexia, and other portal hypertension symptoms. Due to occasional fatal gastrointestinal hemorrhaging, a timely and accurate diagnosis and treatment are particularly important. To improve the understanding of superior mesenteric arteriovenous fistula, we report the clinical data and treatment of a patient with SMAVF and review the relevant literature to analyze the clinical manifestations, imaging features, diagnosis, and treatment. The diagnosis of the disease mainly relies on abdominal enhanced CT+3D reconstruction or digital subtraction angiography, and the treatment is mainly vascular interventional fistula occlusion. So, improving the understanding of the disease can lead to early detection and early treatment and help avoid misdiagnoses.

Statistical methods

The statistical analysis was conducted using SPSS 21.0. GraphPad Prism 8 was used to draw the graphs. The measurement data were expressed as the mean ± SD. The count data were expressed as cases (percentage) [n (%)].

Case presentation

A 17-year-old female who had suffered from diarrhea and abdominal distension for ten days was admitted to the Department of Gastroenterology of the Second Affiliated Hospital of Hainan Medical University. Her stools were yellow, water-like, and she had bowel movements 10-20 times a day, about 30-100 ml each time. There was no mucus, pus, or blood in her stools and there were no severe sensations after the internal urgency. The patient had no chills, fever, or yellow staining of the skin and sclera. The patient had been treated in a local hospital with antibiotics (the specific medication is unknown), but her condition did not improve. One and a half years ago, the patient suffered abdominal, head, face and limb injuries caused by a car accident. Under general anesthesia with tracheal intubation, the patient underwent “laparotomy, liver rupture repair, cecum wall rupture repair, [and] right abdominal wall rupture repair”. The opera-

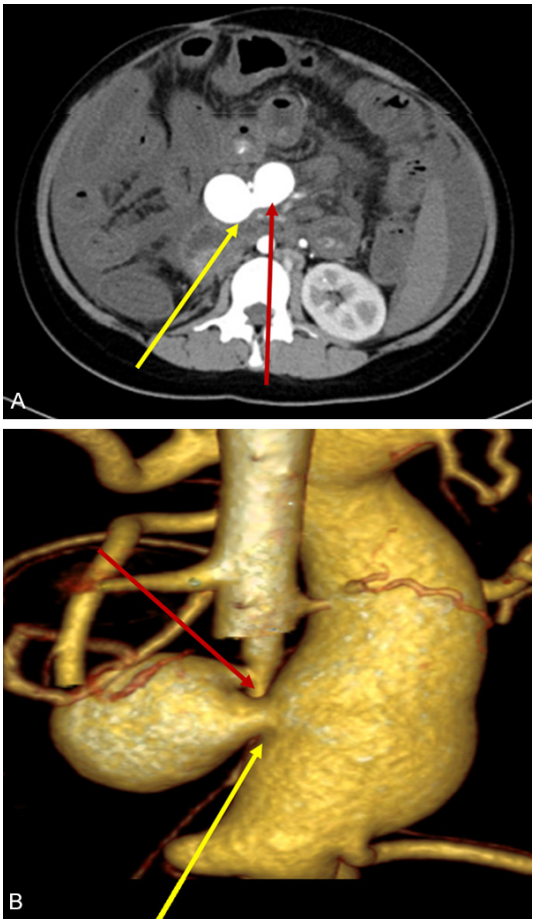

Figure 1. A. CT enhancement shows the formation of a pseudoaneurysm of the superior mesenteric artery (red arrow) in the arterial phase, the early development of the superior mesenteric vein and the high dilation (yellow arrow); B. VR revascularization showed a clear communication between the superior mesenteric artery pseudoaneurysm neck (red arrow) and the superior mesenteric artery pseudoaneurysm and the superior mesenteric vein (yellow arrow).

tion was successful and the patient recovered well after the operation. Physical examination: T 37.1 degrees centigrade, P 78 times/min, R 20 times/min, BP 146/86 mmHg, flat abdomen, on a symmetrical, mid-upper abdomen a 20 cm long surgical scar can be seen, soft abdomen, upper abdomen, subxiphoid tenderness, no rebound pain, and no vascular murmur. Her liver and spleen were not touched. Her intestinal sounds were active, about 12 times per minute. Laboratory results: WBC 13.16 × 10^9/L, HGB 109 g/L, PLT 167 × 10^9/L, N% 89.0%, absolute number of neutrophils 11.71 × 10^9/L, C-reactive protein 96.52 mg/L. Stool routine: color, brown-yellow, character, water-like stool, red blood cells, 20-25/HP, white blood cells, 1-3/HP, occult blood test, positive (3+); no abnormal stool culture, urine sediment: transparency, clearance, urine protein, 1 (++) g/L, urine vitamin C, 0.4 (++) g/L, crystallization, 142.03 (+++++) per u. After admission, the patient was given a liquid diet, antibiotics including cefoperazone sodium, tazobactam sodium, and tinidazole, *Bifidobacterium* quadruple live bacteria tablets to regulate here intestinal flora and symptomatic support treatment. The patient's condition gradually worsened, and the number of her diarrhea bowel movements increased. The patient underwent abdominal enhanced CT+3D reconstruction on 15 August 2017 (**Figure 1**). The diagnosis of superior mesenteric arteriovenous fistula was confirmed based on the patient's history and symptoms. A superior mesenteric arteriography and a stent placement of the superior mesenteric artery were performed on 16 August 2017 (**Figure 2**). Surgical procedure: 1. The Seldinger technique punctured the left brachial artery successfully and then we inserted a sheath tube. A pig catheter was then inserted using the catheter guide wire technique for an abdominal aortography. A pseudoaneurysm of the superior mesenteric artery (about 3 cm in diameter) could be seen about 5-6 cm away from the opening of the superior mesenteric artery. It communicates with the superior mesenteric vein and shows the same image of the superior mesenteric artery during the arterial phase. The aneurysm and superior mesenteric vein portal vein were developed simultaneously, and the superior mesenteric vein and portal vein were significantly dilated. 2. The inferior catheter entered the superior mesenteric artery angiography and clearly showed the rupture of the aneurysm. The hard exchange wire entered the distal superior mesenteric artery and the heparinization (25 mg). The Gore covered stent (viahban, 5 mm × 5 cm) entered the superior mesenteric artery, then it located and released the artery. The angiography showed that the aneurysm was isolated, the stent was in a good position, and the mesenteric artery aorta was in a good position. The branches were retained, the catheter guide sheath was removed, the hemostasis was compressed, the blood loss was 2 ml, and the operation went smoothly. The patient recovered well and was discharged from the hospital. After her discharge, she was treated with rivaroxaban (15 mg per day) for a long time. The patient was followed up for 2 years and recovered well.

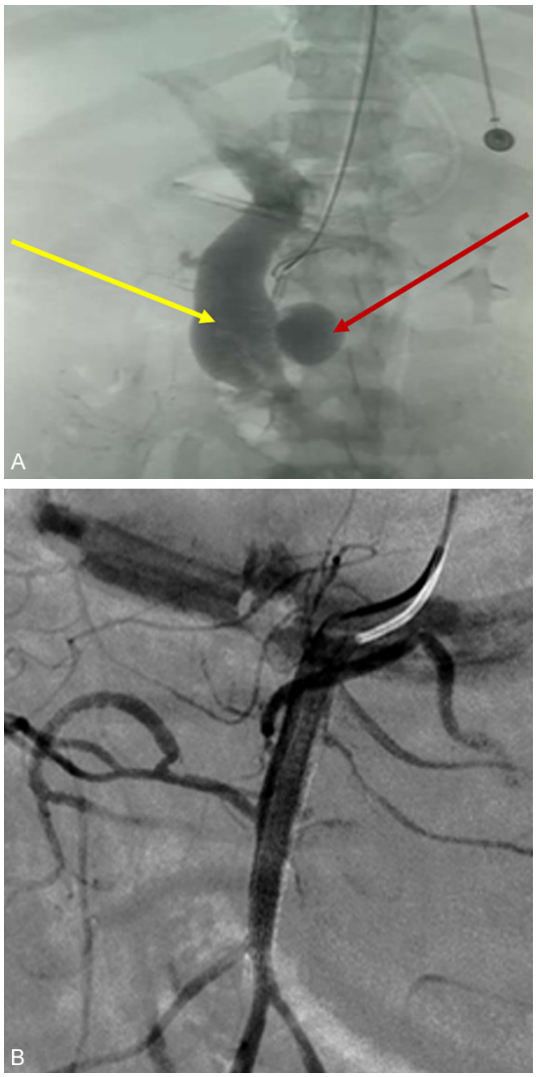

Figure 2. A. The DSA showed a pseudoaneurysm formation (red arrow) of the superior mesenteric artery using superior mesenteric artery angiography. The superior mesenteric artery was well developed and highly dilated (yellow arrow); B. The pseudoaneurysm disappeared after the stent-covered superior mesenteric artery implantation.

The patient was informed about of this study and signed the informed consent forms. This study was approved by the Second Affiliated Hospital of Hainan Medical University.

Discussion and conclusion

Superior mesenteric arteriovenous fistula is very rare clinically. To date, there have been 34 reports. The causes, clinical manifestations, signs, radiological findings, therapeutic means, and outcomes are summarized in **Table 1**. The first case of superior mesenteric arteriovenous fistula was reported by Movitz and Finnel in the 1960s [1]. The disease is caused by trauma or iatrogenic injuries [2, 3], such as a stabbing, an ileectomy, a right hemicolectomy, and on the like, which lead to direct arteriovenous communication, and has the clinical manifestations of abdominal pain, abdominal distention, diarrhea, gastrointestinal bleeding, congestive heart failure, etc. [4]. The main clinical manifestation of this case was acute diarrhea. At present, how diarrhea is caused by a superior mesenteric arteriovenous fistula is unknown. The pathogenesis of diarrhea may be related to the occurrence of regional portal hypertension due to the direct entry of arterial blood flow into the vein through the fistula orifice after the formation of the arteriovenous fistula. Due to long-term portal hypertension, intestinal congestion, edema, erosion, and an increased permeability of the intestinal blood vessels lead to intestinal barrier damage to the intestinal cells, resulting in decreased secretions of the digestive enzymes, a reduced absorptive capacity, secretory dysfunction, and an imbalance of the normal intestinal microflora, leading to diarrhea [5-8]. For diarrhea patients with an elevated portal vein pressure, after the placement of a transjugular intrahepatic shunt, the patients' portal vein pressure decreases, and the diarrhea is significantly alleviated [9].

There are two types of superior mesenteric arteriovenous fistula, the U-type and the H-type. The U-type occurs when the superior mesenteric artery or its branches directly communicate with the veins and is mostly iatrogenic. The H-type occurs when the superior mesenteric artery or its branches communicate with the veins through pseudoaneurysms and is mostly traumatic [10]. Superior mesenteric arteriovenous fistula (SMAVF) is a relatively insidious disease and is difficult to diagnose clinically because there are few positive signs in the clinical examination. Abdominal vascular murmurs and ascites are the most common signs [11].

The diagnosis of SMAVF mainly relies on imaging. Abdominal ultrasonography is helpful in the diagnosis of this disease. Color Doppler flow imaging (CDFI) can detect blood flow changes sensitively. The arterialization of the intravenous blood flow spectrum is characteristic and can be used as the preferred screening method [12]. Early imaging of the superior mesenteric

Table 1. A summary of the superior mesenteric arteriovenous fistula cases

Case	Sex	Age	Cause		Clinical manifestation					Sign			Diagnostic means		Therapeutic means		outcome	
			Surgical history	Trauma history	Abdominal pain	Abdominal distension	Haematemesis	Bloody stool	Diarrhea	Vascular murmur	Ascites	Tremor	CT vascular reconstruction	Angiography	Vascular intervention	Surgery	Good	Death
Taylor, et al [18], 1965	F	43	√							√				√		√	√	
Spellman MW, et al [19], 1967	M	32		√	√					√						√	√	
Varner JE, et al [20], 1969	M	20		√	√					√				√		√	√	
UflackerR, et al [21], 1982	M	41	√		√				√	√				√	√		√	
Rosenthal D, et al [22], 1987	M	16		√	√									√		√	√	
Donell ST, et al [23], 1988	F	53	√							√						√	√	
Kato S, et al [24], 1993	M	63	√				√					√		√		√	√	
Khan TFT et al [25], 1999	M	26	√		√											√	√	
Chen YC, et al [11], 2000	F	41	√		√								√	√		√	√	
Mick SL, et al [26], 2003	F	62	√		√				√	√			√	√	√		√	
J. Chiriano, et al [27], 2005	F	20		√						√		√		√		√	√	
C.-E. Xu, et al [28], 2006	F	43	√			√	√	√			√			√		√	√	
KK Wong, et al [14], 2007	F	67	√								√		√		√		√	
CG Wu, et al [2], 2008	M	20		√	√					√				√	√		√	
J. Chiriano, et al [29], 2009	M	46	√	√	√					√				√	√		√	
Narayanan G, et al [30], 2008	F	24	√	√								√		√	√		√	
Yeo KK, et al [31], 2008	F	39	√	√	√					√			√	√	√		√	
Weinstein D, et al [4], 2009	F	23	√	√	√								√	√	√		√	
Lau KY, et al [32], 2009	M	50	√				√	√					√	√	√		√	
J.P. Eiberg, et al [33], 2009	M	42	√		√				√					√	√		√	
White RD, et al [34], 2010	M	60	√						√					√	√		√	
Shintani T, et al [35], 2011	M	37	√		√				√					√	√		√	
Lingjun Liu, et al [36], 2012	M	59	√		√				√				√		√		√	
Popović DDj et al [17], 2012	M	49	√		√	√				√		√	√	√		√	√	
Temin NN, et al [37], 2012	F	45	√					√						√	√		√	
HusseinM, et al [10], 2013	M	61	√		√									√	√	√	√	
An T,et al [38], 2013	M	35	√		√		√	√	√	√			√	√	√		√	
Zhao Y, et al [39], 2014	M	59	√		√				√				√	√	√			
Grujić D, et al [40], 2015	M	45	√					√	√				√					√
Ahmad Sayed Awad, et al [41], 2016	M	24	√		√								√	√	√		√	
ZY Xie, et al [13], 2016	M	51				√	√	√			√		√	√	√			
Alparslan Kılıç, et al [42], 2017	M	44	√	√	√	√				√				√	√		√	
Lucy D. Miller, et al [43], 2018	M	30		√	√									√		√	√	
Zhao Y, et al [44], 2018	M	34	√				√	√		√				√	√		√	

artery, venous malformations, or a portal vein system during the arterial phase on abdominal enhanced computed tomography may indicate the disease. Abdominal CT angiography and three-dimensional angiography are the most reliable ways to diagnose the disease [11]. The CT reconstruction technique is the best method for the noninvasive diagnosis of this disease. Multiplanar reconstruction and curve planar reconstruction can display malformed blood vessels on the same plane and show the relationship between the fistula and the surrounding tissues. Maximum intensity projection can observe from multiple planes and angles, showing the anatomical structure more precisely. Volume rendering (VR) can display three-dimensional images of the diseased vessels, and it has a strong stereoscopic sense [13]. Digital subtraction angiography (DSA) is the gold standard for the diagnosis of superior mesenteric arteriovenous fistulas. It can dynamically reflect the morphological information of the blood vessels, determine the location and size of the fistula and the severity of the portal hypertension [11].

There are several methods of visualizing an SMAVF. Abdominal ultrasonic scanning, CT, or MRI usually are the first to reveal a vascular deformity [14]. DSA is the gold standard for defining the exact anatomical location and the extent of mesenteric vessel involvement. However, this procedure is typically used only for treatment. So CTA and CTV are widely used clinically.

There are two main treatments for superior mesenteric arteriovenous fistula: 1. Surgery: Surgical ligation of the fistula of the arteriovenous fistula or the removal of the jejunum seriously affected by varicose veins is generally used as a second-line remedy after the failure of vascular interventional surgery due to the great trauma. 2. Vascular interventional fistula occlusion: interventional coil embolization or covered stent occlusion of the fistula [15]. With the development of interventional technology and the improvement of embolic materials, embolization has become the preferred treatment for superior mesenteric arteriovenous fistula [16], and the patients can recover completely after their operations [17]. The patient has no symptoms such as abdominal pain, abdominal distension, diarrhea, or other complications after the interventional embolization with a covered stent. No complications such as coil displacement or portal vein thrombosis were found. The patients were followed up for two years and recovered well.

The patient had a clear history of trauma and abdominal surgery. The first symptoms were regional portal hypertension such as abdominal distension and diarrhea. The diagnosis was confirmed by enhanced CT and VR vascular reconstruction. We believe that for patients with a previous history of trauma or abdominal surgery, if there is intractable diarrhea, abdominal distension, abdominal pain, or other symptoms of regional portal hypertension, we should pay attention to the possibility of a superior mesenteric arteriovenous fistula. It is necessary to check in detail whether the patient has an abdominal vascular murmur or ascites and do an abdominal enhanced CT or DSA examination as soon as possible to make a definite diagnosis. Interventional treatment is the first choice for superior mesenteric arteriovenous fistula. The effect is very significant. If the interventional therapy is ineffective, surgery can be performed to treat the fistula with a ligation of arteriovenous fistula.

However, the research also has some limitations., The experimental data are not sufficiently inclusive. Therefore, extensive research should be conducted to supplement our study.

Acknowledgements

This work was supported by the Hainan Natural Science Foundation Youth Project (819QN360).

Disclosure of conflict of interest

None.

Abbreviations

SMAVF, Superior mesenteric arteriovenous fistula; CT, computed tomography; 3D, three dimensional; DSA, digital subtraction angiography; VR, Volume rendering.

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