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CASE REPORT

Inferior mesenteric arteriovenous fistula with colonic ischemia: a case report and review of the literature

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Abstract
Inferior mesenteric arteriovenous fistula is a rare abnormal high flow communication with only 40 primary and secondary cases reported in literature. Shunting of arterial flow through the inferior mesenteric vein to the portal system can cause a variety of nonspecific clinical signs and symptoms usually associated with the diagnosis of arteriovenous malformation. Symptom intensities are flow-dependent and can range from minimal abdominal symptoms to severe heart failure due to left to right shunt. We report the case of a 72-year-old man without past history of abdominal surgery or trauma who was referred to our department for a 2-month history of intermittent diarrhea and abdominal pain caused by an arteriovenous fistula involving the left colic artery and the inferior mesenteric vein. A progressive and spontaneous improvement of symptoms and a control CT scan that confirmed the reduction of venous vascular engorgement and regression of parietal thickening of the left and sigmoid colon permitted a non-operative management.
Inferior mesenteric arteriovenous fistula can be a rare cause of ischemic colitis and, if necessary, an appropriate treatment based on high clinical suspicion can reduce the risk of complications related to a missed diagnosis.

Keywords Inferior mesenteric arteriovenous fistula · Ischemic colitis · Arteriovenous malformation · Portal hypertension

Abbreviations

IMA-V	Inferior mesenteric artery and vein
AVF	Arteriovenous fistula
CT	Computed tomography

Introduction

Inferior mesenteric artery and vein (IMA-V) arteriovenous fistula (AVF) is a rare abnormal high flow communication with only 40 primary and secondary cases reported in literature (Tables 1 and 2). It is characterized from nonspecific clinical signs and symptoms such as abdominal pain, gastrointestinal bleeding, ischemic colitis, portal hypertension, and heart failure. Primary or congenital AVFs occur from undifferentiated embryonic vessels that fail to differentiate into arteries and veins. Secondary IMA-V AVFs can occur following penetrating abdominal injuries, arterial catheterization, or surgery [1].

Symptom intensities are flow-dependent and can range from minimal abdominal symptoms to severe heart failure due to left to right shunt. Furthermore shunting of arterial flow through the IMV to the portal system can cause portal hypertension that is usually associated with the diagnosis of AVF. According to its infrequency and great variability of symptoms, a radiological or intraoperative examination is often necessary to reach the diagnosis of IMA-V AVFs. Similarly, as regards treatment, it should be adapted to clinicopathological characteristics associated with this rare pathologic process.

Herein we present a rare case of transitory colonic ischemia caused by a congenital IMA-V AVF. At our knowledge, this is the 4th case reported in literature that was managed conservatively. We also review the literature and discuss the characteristics and treatments of this rare pathologic process.

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Table 1 Congenital cases of inferior mesenteric arteriovenous fistula

Case	Sex	Age	Management
Van Way et al. 1971	F	72	Left hemicolectomy
Sabatier et al. 1978	M	22	None (patient's refusal)
Oyama et al. 1980	M	70	Left hemicolectomy
Manns et al. 1990	M	33	Left hemicolectomy; mesenteric vein-caval anastomosis
Baranda et al. 1996	M	63	Ligation and resection of fistula
Capuano et al. 2004	F	76	Embolization; ligation of AVF
Nemcec and Yakes 2005	M	65	Vein embolization
Matsui et al. 2007	M	N/A	Embolization
Kim et al. 2008	M	56	Percutaneous portal angioplasty; left hemicolectomy
Metcalf et al. 2008	M	50	Left hemicolectomy
Türkvatan et al. 2009	M	83	Left hemicolectomy
Akgun et al. 2013	M	48	Total colectomy
El Muhtasaeb et al. 2013	M	57	Embolization; left hemicolectomy
Plotkin et al. 2013	F	70	Embolization
Takahashi et al. 2013	M	78	Resection
Justaniah et al. 2013	M	43	Embolization
Athanasiou A et al. 2014	M	63	Embolization; left hemicolectomy
Brucher et al. 2014	M	59	Arterial embolization
Noor et al. 2016	M	61	Left hemicolectomy
Cheng et al. 2017	M	62	Left hemicolectomy
Lee et al. 2017	M	56	Left hemicolectomy
Hendy et al. 2018	M	24	Embolization
Das Gupta et al. 2019	M	46	Embolization
Rossi et al. 2020	F	63	None (patient's refusal)
kunioka et al. 2020	M	57	Conservative management
Current case	M	72	Conservative management

Table 2 Traumatic–iatrogenic cases of inferior mesenteric arteriovenous fistula

Case	Sex	Age	Management
Houdard et al. 1970	F	51	Left hemicolectomy
Hirner et al. 1978	F	43	Left hemicolectomy
Capron et al. 1984	F	60	Intraarterial embolization
Slutski et al. 1988	M	63	Embolization
Peer et al. 1989	M	63	Embolization
Pietri et al. 1990	M	62	Left hemicolectomy
Pietri et al. 1990	F	60	Intraarterial embolization
Vauthey et al. 1997	M	58	Percutanous embolisation
Okada et al. 2002	F	69	Left sigmoidectomy
Fabre et al. 2005	M	N/A	Embolization
Gorospe et al. 2012	M	58	Embolization; total colectomy
Jeon DO et al. 2013	F	53	Surgical excision without colectomy
Carbonell et al. 2014	M	66	Embolization
Charalambous et al. 2020	F	73	Embolization
Doi et al. 2020	M	81	Embolization

Case report

A 72-year-old man without past history of abdominal surgery or trauma was referred to our department for a 2-month history of abdominal pain and intermittent diarrhea, without bloody stools. The physical examination of the abdomen was unremarkable and the patient had no signs or symptoms of portal hypertension. Full laboratory tests were also unremarkable. The colonoscopy showed an edematous and fragile mucosa with mild subepithelial hemorrhages. These findings were consistent with a diagnosis of ischemic colitis extended from the rectosigmoid junction to the splenic flexure. Biopsies taken at these levels showed submucosal hemorrhage and edema with few areas of fibrosis and were consistent with the diagnosis of ischemic colitis. A computed tomography (CT) of the abdomen confirmed a bowel wall thickening with congestion and decreased enhancement in the same segments associated to dilation of the vascular branches of the IMA-V plexus (Fig. 1a–b). A multidetector CT angiography of the IMA was performed and demonstrated an AVF involving the left colic artery and the inferior

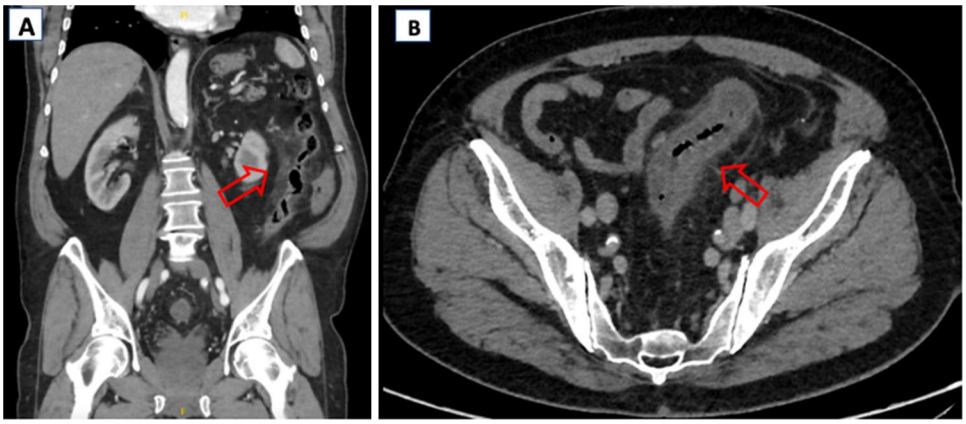

Fig. 1 Abdominal CT scan showing a bowel wall thickening with congestion and decreased enhancement extended from the rectosigmoid junction to the splenic flexure. **a** Coronal and **b** axial view

mesenteric vein. The IMV appeared dilated and varicose with a caliber of 18 mm and with a fast wash-out in portal system. (Fig. 2a–b). Colonic ischemia was the result of the arterial flow shunting through the inferior mesenteric vein and portal system with a venous stasis caused by the AVF.

Initially, we considered embolization of the feeding artery with possible subsequent colic resection; however, in the following month, the patient reported a progressive and spontaneous improvement of symptoms. An angio CT scan with color subtraction imaging permitted to optimize the vascular study and confirmed the reduction of venous vascular engorgement due to venous collateral circles development with significant regression of parietal thickening of the left and sigmoid colon (Figs. 3 a–b, 4 a–b). Although of considerable interest for the completeness of the clinical case, these findings made it possible to avoid the execution of a control angiographic examination. Resolution of colonic ischemia confirmed endoscopically and the complete symptoms remission, verified at 1-year follow-up, permitted a non-operative management.

Discussion

Different etiopathogenesis have been identified for the development of IMA-V AVFs. Congenital AVFs occur from undifferentiated embryonic vessels that fail to differentiate into arteries and veins.

Secondary or iatrogenic IMA-V AVFs can occur following blunt or penetrating abdominal injuries, arterial catheterization, cholangiography, splenoportography or various surgical procedures such as a left hemicolectomy or sigmoidectomy [1]. In literature, it is also reported that the rupture of a congenital arterial aneurysm very close to a vein can also result in the formation of an AVF [2]. Our patient had no medical history of abdominal surgery or trauma; therefore, was possible to assume that the fistula originated from idiopathic etiology. An AVF is usually associated with decreased arterial blood flow to the tissue beyond the fistula and increased venous pressure distal to the fistula.

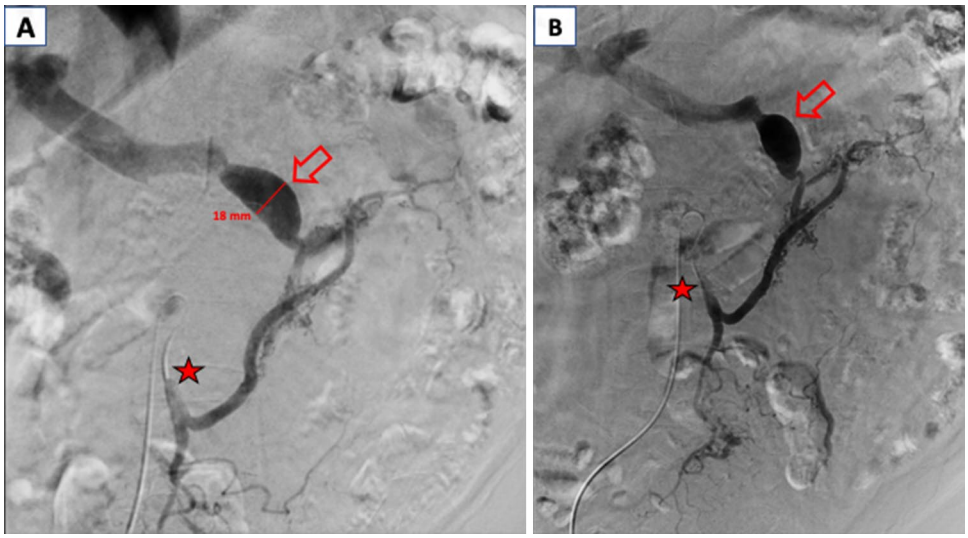

Fig. 2 **a**, **b** Main findings of multidetector CT angiography. The inferior mesenteric vein appeared dilated (red arrow) and supplied by inferior mesenteric artery (red star)

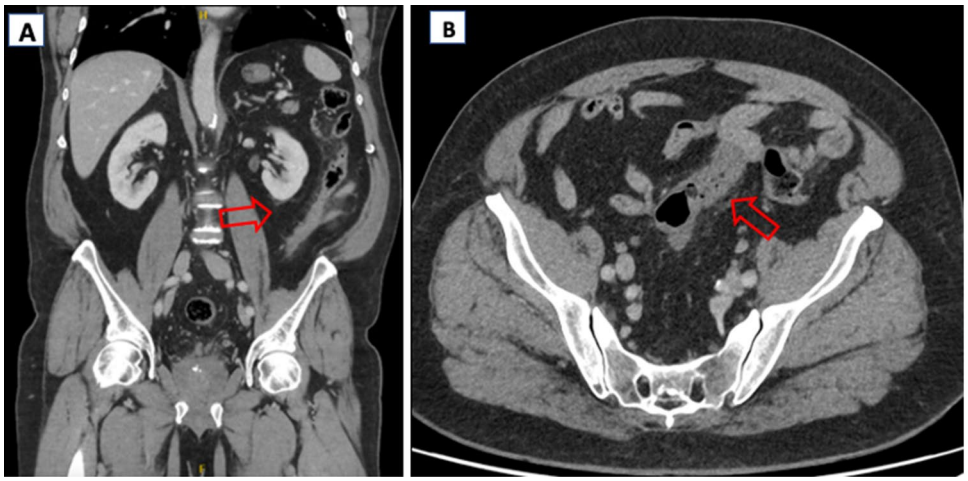

Fig. 3 Control CT scan showing the reduction of venous vascular engorgement with significant regression of parietal thickening of the left and sigmoid colon. **a** Coronal and **b** axial view

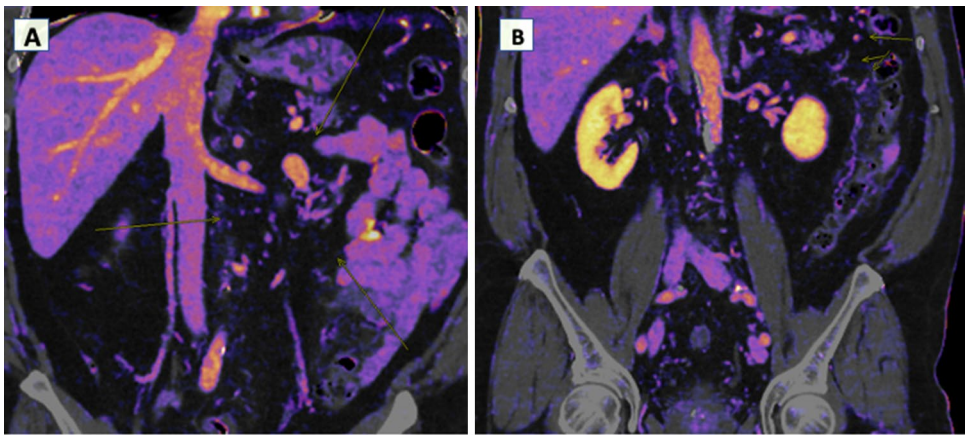

Fig. 4 Angio CT scan with color subtraction imaging showing a reduction of venous vascular engorgement (**a** coronal view) and a significant regression of parietal thickening of the sigmoid colon (**b** axial view)

Colonic ischemia, commonly associated with this type of arteriovenous malformation, is the result of a steal phenomenon of the arterial flow through the inferior mesenteric vein and portal system. As reported by Slutski S. et al., this vascular engorgement of the bowel mucosa can be also responsible of lower GI bleeding [3]. Portal hypertension with esophageal or duodenal varices can be instead responsible of upper GI bleeding which is often associated with ascites, hepatic encephalopathy, and splenomegaly [4]. The left to right shunt caused by these IMA-V AVFs is the real responsible of the pathophysiologic alteration of bowel function and of the variety of nonspecific clinical signs and symptoms related. High flow comunication through an AVF, due to left to right shunt, can lead to cardiac failure that is another important symptom of these arteriovenous malformations. As reported by Fabre et al. the embolization of the AVF can also result in improvement of cardiac function and an increased ejection fraction [5]. Two major clinical manifestations of IMA-V AVF are portal hypertension and ischemic bowel disease. The only presence of abdominal mass, also reported in literature, is rare and usually leads to missed diagnosis and suboptimal intervention [6]. To prevent misdiagnosis, a high clinical suspicion is requested but for a diagnostic certainty a clinical sign of an ischemic colon combined to the angiographic proof of the fistula's presence is mandatory. Concerning AVF treatment, our literature search found 18 cases of IMA-V AVF that received surgery, 14 embolization, 5 combination therapy, and 3 non-operative management. Two patients of the non-operative management group, who refused intervention, remained paucisymptomatic and were therefore enrolled in a follow-up program (4, 7–10). According to this great variability of symptoms, the treatment of IMA-V AVF remains case specific and can range from non-operative management, percutaneous endovascular embolization of the feeding artery or surgical correction of the AVF with or without bowel resection. A non-operative management for this type of rare pathologic process seems to be feasible only for paucisymptomatic patients when there is no development of portal hypertension and in presence of mild colonic ischemia. Dual arterial and venous embolization with or without subsequent bowel resection may be needed to manage severe portal hypertension secondary to this rare pathologic process. We reported an extremely rare case of

IMA-V AVF that was managed conservatively without any intervention. In our case, we observed progressive and spontaneous improvement of symptoms which, due to venous collateral circles development, was associated to reduction of venous vascular engorgement and significant regression of colic parietal thickening.

IMA-V AVF, bypassing the capillary bed of the colon, can be a rare cause of ischemic colitis. If necessary, an appropriate treatment based on high clinical suspicion can reduce the risk of complications related to a missed diagnosis.

Author contributions AC and VS performed the literature review and wrote the manuscript. BG reviewed and edited the manuscript. All authors approved the submission.

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Declarations

Conflict of interest The authors have no conflicts of interest to declare.

Human rights All procedures followed have been performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki and its later amendments.

Informed consent Informed consent was obtained from the patient for being included in this report.

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