

REVIEW

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Transarterial embolization of secondary inferior and superior mesenteric artery arteriovenous fistulas: a systematic review

Abhinandan Ruge^{1*}, Nidhi Kumar², Lija K. Jose² and Vidya Mahalmani³

Abstract

Background: Acquired arteriovenous fistula are rare complications of a piercing injury, including a gunshot or knife injury, the rupture of a preexisting aneurysm into a neighboring vein, or a complication of arterial catheterization or abdominal surgical techniques, most often colonic resection. Our aim was to develop an online systematic review of the literature to determine all documented incidents of endovascular embolization of secondary mesenteric Arteriovenous Fistula, spanning key databases as well as pertinent publications, from 1982 to 2019. Our case was included as well. The primary objectives were technical success and death; the secondary outcomes were the frequency of complications and the advice of the operators.

Main body: Forty articles, comprising 41 patients, including our own, were reviewed. The superior mesenteric vessels are the most commonly affected by fistulas. Endovascular embolization was used in 80% of the cases. 100% of the patients had technical success. The intraoperative death rate was 0%, with a 90-day mortality rate of 8%, half of which was unrelated to the original pathology. More than 60% of patients who underwent successful treatments had no severe problems. All others had uneventful recoveries, with only 12% experiencing mild problems that were managed conservatively.

Conclusions: The majority of authors supported this therapeutic approach thereby making endovascular glue embolization the treatment of choice for superior mesenteric arteriovenous fistula and inferior mesenteric arteriovenous fistula in high risk patients and in patients with multiple co-morbidities.

Keywords: Embolization, Endovascular, Inferior mesenteric arteriovenous fistula (IMAVF), Superior mesenteric arteriovenous fistula (SMAVF), Transcatheter, Glue

Background

AVF is an aberrant direct link between an artery and a vein that bypasses the capillary bed and can be acquired or congenital [1]. Despite the early report of AVF in 1947, fistula of the superior mesenteric artery (SMA) and inferior mesenteric artery (IMA) are still considered rare entities [2], which are easily misdiagnosed [3]. Splanchnic

AVFs have been recorded in hundreds of cases, primarily between the hepatic, splenic, and superior mesenteric arteries, with only a few examples of superior and inferior mesenteric AVFs reported in the literature [4–14]. Simultaneous occurrence of SMA and IMA fistula is the rarest of the rare in the literature, especially treated with an endovascular approach. None of the reviewed articles in this study described the co-occurrence of SMA and IMA AVFs.

Trauma or intestinal surgery may have occurred recently or in the distant past, spanning between hours to decades [15–21]. The clinical presentation of SMA and IMA AVF is atypical and depends on the size and

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location of the fistula, from asymptomatic to abdominal pain with or without diarrhea, portal hypertension, liver impairment, and even gastrointestinal bleeding [22], causing secondary severe portal hypertension [23].

Multidetector computed tomographic (MDCT) and conventional angiography have developed as an important imaging tool in assessing the mesenteric vasculature. Endovascular therapy techniques are gaining popularity, providing a safer, more efficient approach. There have been some rare case reports of effective endovascular therapy. We were fortunate to see a successful instance of transarterial embolization of iatrogenic SMA and IMA AVE. There are no large case series or controlled trials that can be used to support this therapy method. In order to make any conclusions about this therapy, we did a systematic literature review to investigate published accounts of similarly treated cases.

Main text

Case 1

A 46-year-old male presented with multiple episodes of bleeding and the passage of clots per rectum for a day. The patient has been a known case of EHPVO (Extrahepatic portal venous obstruction) since childhood and had a surgical history of splenectomy 20 years back. On admission, his BP was 90/60 mmHg and his pulse rate

was 92 beats/min. Examination of the abdomen revealed generalized tenderness with no evidence of mass per abdomen, shifting dullness, or bruit. Anemia was discovered in the laboratory. An upper gastrointestinal endoscopy revealed minor esophageal and fundal varices with no current bleeding. Colonoscopy was inconclusive due to poor visual field secondary to bleeding. Sclerotherapy was done for the rectal varices (Fig. 1).

MDCT was performed to determine the extent of the thrombus and the site of active bleeding. MDCT demonstrated diffuse hepatic atrophy with compensatory caudate lobe hypertrophy, thrombosis of the intrahepatic portal vein extending into the retro pancreatic splenic vein, and portosplenic confluence with dilatation of the intrahepatic biliary radicles. There are numerous tortuous gastric, mesenteric, colonic, and jejunal varices, with thrombosis within a few of them. The MDCT also detected a superior mesenteric artery aneurysm that drains into the superior mesenteric vein via a convoluted fistulous tract. Fistulous connections between inferior mesenteric artery branches and inferior mesenteric vein tributaries, as well as aneurysmal dilatations near the fistulous tract's origin, were seen in the arterial phase. Dilated feeding arteries and heavily opacified early draining ectatic veins were also visible. Other CT findings were mild to severe ascites, a complicated kidney cyst,

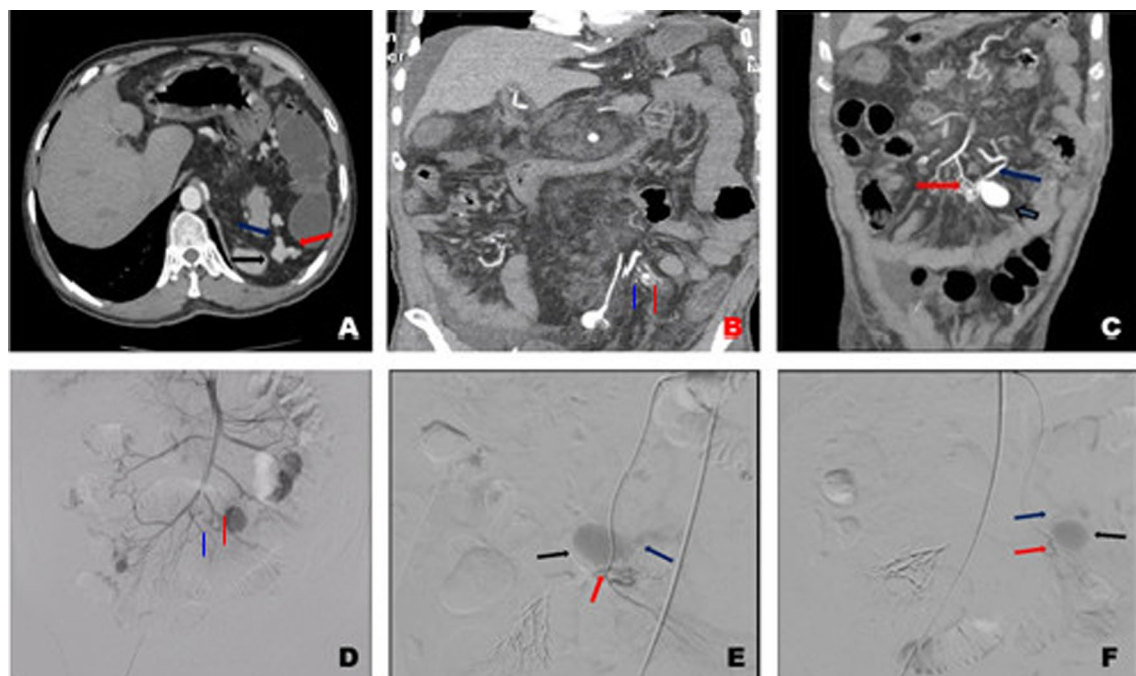


Fig. 1. 46-year-old male, s/p Splenectomy, presented with melena, on CECT, and conventional angiography showing multiple superior and inferior mesenteric arteriovenous malformations. A: showing AVF between SMA & SMV, B, C, D, E, F: showing AVF between IMA & IMV (Red arrow: Artery, Blue arrow: vein, Black arrow: Fistulous arteriovenous malformation)

and a small pleural effusion on the left side. These findings were consistent with those predicted for an arteriovenous fistula caused by SMA or IMA.

Treatment options were explained to the patient along with the high risk of an open surgical invasion because of secondary adhesions and the possibility of massive bleeding from numerous collateral arcades. Hence, the patient was taken for conventional angiography and transarterial glue embolization of AVF. Through the right femoral artery, a celiac, SMA, and IMA angiogram was performed, revealing a normal celiac axis with multiple fistulous malformations of SMA and IMA as focal areas of abnormal blush with multiple collateral arcade formations. Angiogram confirmed the previously reported contrast-enhanced CT findings. Multiple fistulous communications between the distal SMA and SMV, IMA and IMV were confirmed with contrast opacifications of the dilated SMV and IMV documented during the early arterial phase. Using a catheter wire system, the feeding vessels of culprit arcades were selectively cannulated and embolized with 30% glue (*n*-butyl-2-cyanoacrylate-lipiodol combination). The AV fistulas of SMA and IMA were embolized serially under DSA (Digital Subtraction Angiography) guidance. A postembolization angiogram revealed the cessation of opacifications of the aneurysmal malformations and AVF tract.

The patient was strictly monitored for immediate post-embolization syndrome and signs of bowel ischemia. Post-procedure CT showed complete occlusion of the aneurysmal dilatation and fistulous tracts of SMA and IMA. The tortuous varicosities of the prior studies were not visualized in the post-procedure study (Fig. 2).

Data acquisition and methodology

A systematic analysis of the literature was conducted in order to identify all documented examples of endovascular repair of mesenteric AVFs, which are described as abnormal connections between the superior and inferior mesenteric arteries and veins. The authors conducted an exhaustive Web-based review using the terms "Endovascular with inferior mesenteric fistula, superior mesenteric fistula." The recovered literature's references and related articles were also scoped. Articles written in languages other than English have been translated. The online search included all relevant peer-reviewed journal articles published up to September 2019 in the following databases: Elsevier, Wiley online, Springer Link, PubMed, Researchgate, Science Direct, and Google Scholar. The search yielded 40 articles describing 40 individuals with mesenteric AVFs who were treated with various endovascular techniques. In addition to our example, a total of 41 individuals were included in the study (Table 1). The inclusion criteria were case report publications that used a unique endovascular procedure as the primary therapy. Book chapters and systematic reviews, as well as four papers reporting on a hybrid strategy, were excluded from our review (combining endovascular and surgical). Technical success (fistula obliteration with or without problems and without the necessity for open surgical conversion), perioperative, and 90-day mortality were the primary end measures. Procedure-related problems, which are classified as minor and require surveillance or reintervention, as well as the operator's retrospective personal overall improvement, the following therapy was prescribed: The secondary outcome measures were

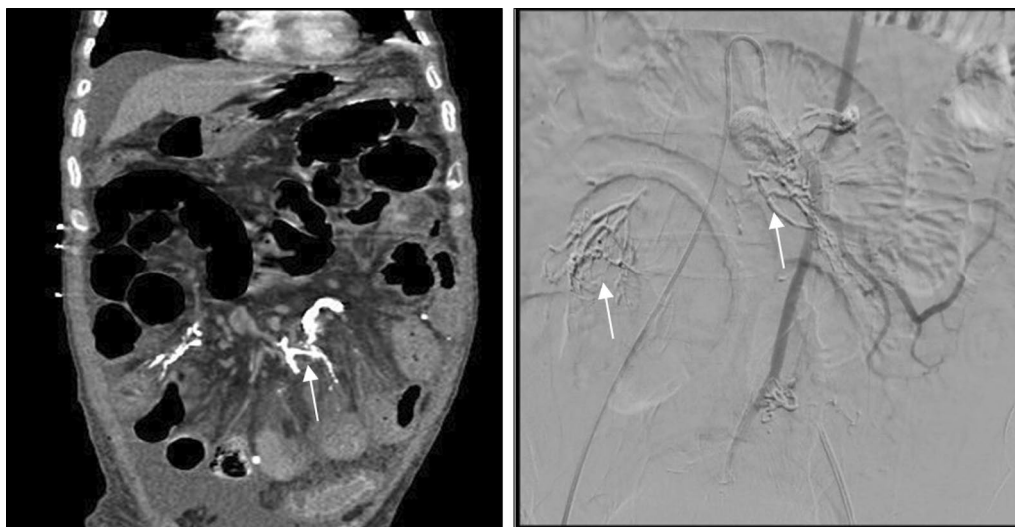


Fig. 2 Post-transcatheter Glue embolization of the secondary SMA & IMA AVF in coronal CECT and DSA images (white arrow)

Table 1 Published case reports of Superior and Inferior mesenteric AVMs treated with an endovascular approach

Authors	No. of pts	Age/sex	Presentation	Site	Procedure	Emergency/elective	Complications	Recommendations
Stephanie Mick et al	1	62/F	Abdominal pain and bloody diarrhea	SMA & SMV	Embolization	Elective	Nil	Favorable than surgical approach
Françaises de radiologie	1	59/M	Pain abdomen	IMA & IMV	Embolization	Elective	Nil	Effective
Khung Keong Yeo et al	1	39/F	Progressive abdominal pain post-surgery	SMA & SMV	Stent graft	Emergency	Nil	Good though long-term results are unknown
Hiong-Ping Hii et al	1	47/F	Abdominal pain and Bruit	Aorta & SMV	Coil embolization	Elective		Alternative to surgical treatment
J. Chiriano et al	1	46/M	Post-traumatic abdominal pain	SMA & PV	Stent graft	Elective	Nil	Safe and effective for short term
Yanping Zhao et al	1	34/M	Melena for 7 days	SMA & SMV	Stent graft	Elective	Stent migration	Safe and effective for short term
Filippo Landi et al	1	47/M	Cirrhosis with hepatitis B & C	SMA & SMV	Embolization	Emergency	Nil	Feasible and safe, alternative to complex surgery
Miguel Nogueira et al	1	64/M	Lower abdominal pain and bloody diarrhea	IMA & IMV	Embolization	Elective	Nil	Case specific and multidisciplinary approach is needed
Lucy D et al	1	30/M	Abdominal pain and diarrhea	SMA & SMV	Stent graft	Emergency	Nil	Combined approach in mesenteric steal syndrome is good
Tomasz Jargiello et al	1	51/M	Nausea and hematemesis	SMA & SMV	Embolization	Elective	Nil	Safe and effective treatment
Lau KY et al	1	50/M	Recurrent hematemesis	SMA & SMV	Coil embolization	Emergency	Nil	Safe, concise, complete and quick life-saving measure
KengoKaai et al	1	66/M	Pain abdomen and bloody diarrhea	IMA & PV	Embolization	Elective	Nil	Combined approach is better in complicated cases
Renan Uflacker et al	1	41/M	Acute abdomen pain	SMA & PV	Coil embolization	Elective	Nil	Safer approach
SaruhanCekirge et al	1	53/M	visceral artery aneurysms	SMA & SMV	Coil embolization	Elective	Nil	Important tool in the treatment of arterio-venous fistulas
Haitham Al-Khayat et al	1	24/F	Gunshot wound abdomen	SMA & SV	Stent graft	Emergency	Nil	Multidisciplinary approach is recommended
Tianzhi An et al	1	35/M	acute hematemesis and melena	SMA & SMV	Coil embolization	Elective	Nil	Safe method with excellent results
Maher Hussein et al	1	61/M	Pain abdomen and bleeding per rectum (hematochezia)	SMA & SMV	Embolization	Elective	Bowel ischemia	Expected complication though rare
AndrasBikk et al	1	36/M	Postoperative pain and tenderness	SMA & SMV	Stent graft	Elective	Nil	Excellent management in complicated cases
Cory M. Fielding et al	1	59/F	Gunshot wound to abdomen	SMA & Duodenum	Coil embolization	Elective	Nil	Best approach
Thorstad BL et al	1	57/M	Hematemesis and melena	SMA & Duodenum	Embolization	Elective	Expired	Important to treat symptoms
Lingjun Liu MD et al	1	59/M	Recurrent abdominal pain and diarrhea	SMA & SMV	Embolization	Elective	Nil	Safer management

Table 1 (continued)

Authors	No. of pts	Age/sex	Presentation	Site	Procedure	Emergency/elective	Complications	Recommendations
J. Deitrick et al	1	16/M	Gunshot wound abdomen	SMA & PV	Coil embolization	Elective	Pancreatic head necrosis, bowel ischemia	Multidisciplinary approach is favorable
Srinivas B Desai et al	1	35/M	Acute bleeding	SMA & SMV	Coil embolization	Elective	Nil	Multidisciplinary understanding
Peter Hendy et al	1	24/M	Abdominal pain, weight loss, bloody diarrhea	IMA & IMV	Embolization	Elective	Nil	Complete cure
Oliver F. Hennessey et al	1	36/M	Pain abdomen	SMV & PV	Coil embolization	Elective	Nil	Good outcome
PezhmanFarshidm-ehr et al	1	32/F	Acute abdominal Pain	SMA & SMV	Coil embolization	Elective	Nil	Successful outcome
Jaideep Das Gupta et al	1	46/M	Severe portal hypertension	IMA & IMV	Embolization	Elective	Nil	Minimally invasive intervention
Govindarajan Narayanan et al	1	24/F	Postoperative for multiple gunshot wounds	SMA & SMV	Stent graft	Elective	Nil	Wider acceptance
Albert A. Nemcek et al	1	56/M	Pain abdomen and black colored stools	IMA & IMV	Embolization	Elective	Nil	Successful outcome
Amir Peer et al	1	63/M	Abdominal pain and bloody stools	SMA & SMV	Coil embolization	Elective	Nil	Good performance
Chun-Gen Wu et al	1	20/M	persistent epigastric pain, vomiting, and nausea, with marked deterioration of his clinical condition	SMA & SMV	Stent graft	Elective	Nil	Effective management
Ronald G. Repasky et al	1	22/M	Abdominal gunshot wound	SMA & SMV	Embolization	Elective	Nil	Effective treatment
David Rosenthal et al	1	16/M	Abdominal pain with pulsatile mass in the epigastrium	SMA & SMV	Embolization	Elective	Nil	Multidisciplinary approach
Tsunehiro Shintani et al	1	37/M	Abdominal pain and Diarrhea	SMA & SMV	Embolization	Elective	Nil	Successful treatment
Nathaniel N. Temin et al	1	45/F	Postoperative ascites	SMA & SMV	Embolization	Elective	Nil	Good outcome
AlparslanKılıç et al	1	44/M	Diffuse abdominal pain, tenderness, fullness, loud continuous bruit	SMA & SMV	Coil embolization	Elective	Nil	Successful alternative to surgical management
HeikoUthoff et al	1	84/M	Diffuse abdominal pain with increasing abdominal girth	SMA & PV	Coil embolization	Elective	Nil	Goodoutcome

Table 1 (continued)

Authors	No. of pts	Age/sex	Presentation	Site	Procedure	Emergency/elective	Complications	Recommendations
Daphna Weinstein et al	1	23/F	Abdominal pain and gastrointestinal bleeding 3 years after an abdominal gunshot wound	SMA & SMV	Coil embolization	Elective	Nil	Alternative to surgery
Richard D. White et al	1	60/M	Abdominal trauma, postoperative diarrhea	SMA & SMV	Embolization	Elective	Nil	Successful procedure
Jeremy Wild et al	1	26/F	Post-traumatic injuries with hemodynamic compromise	SMA & SMV	Embolization	Elective	Nil	Good effort outcome
Abhinandan et al	1	46/M	Bleeding per rectum	SMA & SMV + IMA & IMV	Glue embolization	Elective	Nil	Good alternative in high risk patients with multiple comorbidities

strongly in favor, in favor but with caution, awaiting further recommendations before suggesting the procedure, and no or negative advice. Finally, a comparative subanalysis was conducted to evaluate mortality and complication rates between patients getting elective therapy (defined as a procedure that is scheduled) and patients who require urgent therapy (defined as patients requiring urgent intervention due to a critical state or hemodynamic instability).

Results of data analysis

The average age of the cases examined was 50 years (age range 16–84). There were 31 males (59%) and 10 females (41%) among the 41 patients. The superior mesenteric segment was the site of the fistula in 80.4% of patients (33 of 41), the inferior mesenteric segment in 12.1% (5 of 41), and the superior and inferior mesenteric segments in 2.4% of patients (1 of 41). In 7.3% (3 of 41) of patients who had successful treatments, complications occurred. None of the patients had major problems. The symptomatic strategy was beneficial in managing those problems. All minor issues were handled with caution. In reflection, the authors were mainly in favor of endovascular treatment. The therapy modality received strong support from 41 authors, including us. Five writers were enthusiastic, but cautioned that the therapy should be used with caution and in conjunction with a multidisciplinary approach.

Interpretation and discussion

Splanchnic AVFs can be found in a variety of places. The celiac artery or its branches, particularly the hepatic

(45%) and splenic (30%) arteries, are responsible for the majority of cases. In decreasing order of frequency, the superior mesenteric, gastroduodenal, and inferior mesenteric arteries are involved [1, 16, 19]. AVF involving the superior and inferior mesenteric vessels is uncommon in only about 41 cases collectively in the literature [3–14]. SMA and IMA AVFs are abnormal communication between the SMA, SMV, and IMA, IMV, respectively. SMAVFs are most commonly caused by a traumatic event, such as a surgical colon resection. The hemodynamics of the AVF is most likely to blame for the substantial difference in the time it takes for mesenteric AVFs to manifest clinically. A large and rapid flow of blood from the arterial to the venous circulation characterizes high-flow fistulas [14]. According to one study, the duration between surgery and the beginning of clinical SMAVF can be as long as 25 years [24]. Our patient had a history of splenectomy 20 years ago.

The intestine's arteriovenous fistula is a rare cause of gastrointestinal hemorrhage. Gastrointestinal bleeding can occur from a variety of sources. The origins of bleeding in Pietri et al's [19] series included esophageal or cardiac varices due to portal hypertension, massive hematochezia due to cecal AVF rupture, hemobilia due to hepatic biopsy, and bloody diarrhea due to colonic ischemia. The most common sign of mesenteric AVFs is increasing stomach pain, which may be accompanied by malabsorption symptoms [25]. Untreated fistulas, despite the usual delayed and inconspicuous clinical manifestation of SMA and IMA AVF, can have fatal consequences, with a reported mortality rate of up to 25% [26]. Certain

instances of mesenteric AVFs are asymptomatic for a long time before presenting with symptoms. PHTN (Portal hypertension) develops over time when blood is shunted from the arterial to the venous systems, resulting in refractory ascites and severe GI bleeding [25].

Abdominal ultrasound scanning, CT, or MRI are usually the first to reveal vascular deformity [23]. MDCT angiography has recently gained popularity as a method for assessing the mesenteric vasculature. Because of its superior spatial resolution and faster acquisition periods, it allows for the imaging of normal vascular anatomy and gives reliable diagnostic information, allowing assessment of the peripheral visceral branches [27–29]. The gold standard for determining the precise anatomic site and amount of mesenteric vascular involvement is DSA. However, this operation is usually reserved mainly for treatment purposes.

SMAVF surgical ligation is a well-known therapeutic option. However, due to problematic bowel mobilization and adhesions from previous abdominal surgery, the operation can be technically tough. Surgical procedures have a 18% fatality risk [27]. Percutaneous endovascular embolization of the feeding artery is becoming more popular as an option to surgery [17, 19, 21]. It's less invasive, and it's becoming more popular for occluding mesenteric AVFs [30]. Occlusion of AVFs has been accomplished with metal coils, stent-grafts, amplatzer vascular plugs, Onyx, and detachable coils with *N*-butyl-2-cyanoacrylate glue [31]. Recently, percutaneous endovascular treatment of such AVFs has been increasingly performed, using covered stents or embolization [30, 32, 33]. Metallic coil embolization could be problematic when the fistula's flow rate is high and its diameter is greater than 8 mm because of the likelihood of migration into the portal venous system [34]. Embolization may be challenging in situations with congenital or multiple iatrogenic AVFs, as it is only effective if all active and quiescent shunts are closed.

Conclusions

Finally, we provide a case of upper GI bleed caused by several superior and inferior mesenteric AVFs, which was easily and consistently visualized by MDCT and validated by traditional angiography. In the case of suspected mesenteric arteriovenous malformations, we recommend MDCT angiography as the primary study. For the evaluation of possible interventions, conventional angiography can be used. AVF can be misdiagnosed because the patients' unique clinical symptoms are typically nonspecific, and physical examination may not indicate the existence of abdominal bruit. A differential of arteriovenous malformations should be evaluated in the presence of secondary indications of portal hypertension and a history of abdominal surgery. Endovascular embolization

can be safely performed with outstanding results. Endovascular glue embolization (*n*-butyl-2-cyanoacrylate and lipiodol combination) can be the choice of endovascular intervention in high flow arterio-venous malformation and in patients with multiple co-morbidities or high risk patients. It is also an economical option, thus making it the treatment of choice even for patients of lower economic strata and in low socio-economic countries as well.

Abbreviations

AVF: Arteriovenous fistula; CT: Computed tomography; DSA: Digital subtraction angiography; EHPVO: Extrahepatic portal venous obstruction; GI: Gastrointestinal; IMAVF: Inferior mesenteric arteriovenous fistula; IMA: Inferior mesenteric arteriovenous; IMV: Inferior mesenteric vein; MRI: Magnetic resonance imaging; MDCT: Multidetector computed tomographic; PHTN: Portal hypertension; SMAVF: Superior mesenteric arteriovenous fistula; SMA: Superior mesenteric arteriovenous; SMV: Superior mesenteric vein.

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Author contributions

ACR, NK, LKJ and VM were involved in conceptualization. ACR, NK and LKJ were involved in methodology. ACR, NK and VM were involved in formal analysis. ACR and LKJ were involved in writing—original draft. NK was involved in writing—review and editing and performed supervision. LKJ and VM were involved in the investigation and collected resources. LKJ was involved in writing—review. VM was involved in data curation. All authors have read and approved the manuscript.

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Availability of data and materials

Data sharing isn't necessary because all of the data is publicly available and no new data was collected or evaluated.

Declarations

Ethics approval and consent to participate

Not applicable.

Consent for publication

Not applicable.

Competing interests

The authors have no competing interests to disclose.

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