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Case Report

Trans-arterial embolization of a Yakes type IIb inferior mesenteric arteriovenous malformation: A case report and literature review for angio-architecture analysis[☆]

Bo-Ying Su, MD^a, Yen-Chi Wang, MD^b, Mei-Jui Weng, MD^c, Matt Chiung-Yu Chen, MD^{d,*}^a Department of Radiology, Taipei Hospital, Ministry of Health and Welfare, New Taipei, Taiwan^b Department of Radiology, Tri-Service General Hospital, New Taipei, Taiwan^c Department of Radiology, Kaohsiung Veterans General Hospital, Kaohsiung, Taiwan^d Department of Interventional Radiology, Yuan's General Hospital, No.162, Cheng-gong 1st Rd., Lingya District, Kaohsiung City 802, Taiwan

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ABSTRACT

We herein report a case of acute, massive lower gastrointestinal bleeding caused by Yakes type IIb inferior mesenteric arteriovenous malformation, which was successfully treated with endovascular embolization. The Yakes arteriovenous malformation classification provides curative treatment strategies based on specific angioarchitecture, thus serving as a valuable guide during treatment planning. We reviewed reported cases from 1988 to 2022 and conducted an angioarchitecture analysis based on the Yakes classification. We analyzed these reported cases to estimate the treatment success rates of surgery and embolization.

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Introduction

Inferior mesenteric artery arteriovenous malformation or fistula (IMA AVM/AVF) is rare, and even rarer when causing ischemic colitis combined with lower gastrointestinal bleeding. Surgery and embolization are treatment options, but the

success rates of these 2 treatment options based on reported cases to date are lacking. The Yakes AVM classification system provides curative treatment strategies based on specific angioarchitecture [1], thus serving as a valuable guide during treatment planning.

Considering the clarity of available images for angioarchitecture analysis, we conducted a literature review of PubMed

Abbreviations: CT, computed tomography; IMA AVM/AVF, Inferior mesenteric artery arteriovenous malformation or fistula; IMV, Inferior mesenteric vein; TA, Trans-arterial route; TV, Transvenous route.

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* Corresponding author.

E-mail address: jjychen@gmail.com (M.C.-Y. Chen).

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Table 1 – Patients enrolled for angioarchitecture analysis.

Case number	Author and publication date	Age and gender	AVM type	Surgery or embolization	Route	Treatment outcome	Treatment complications
1	Nogueira 2018	64 male	Ia/b	Embolization	TV	Success	None
2	Pakray 2021	76 male	Ia/b	Embolization	TA	Success	None
3	Coulier 2018	45 male	Ia/b	Embolization failure with bailout surgery	TA	Success	None
4	Gelonch 2017	73 male	IV	Presurgery embolization	NA	Success	None
5	Shiraishi 2022	70 male	IV	Surgery	NA	Success	None
6	Jubashi 2021	57 male	Ia/b	Embolization	TV	Success	None
7	Justaniah 2013	43 male	Ia/b	Embolization	TV	Success	None
8	Hong 2021	64 male	IIIb	Embolization	TA	Success	None
9.	Plotkin 2013	71 female	IIIa	Embolization	TV	Success	None
10.	Nemcek 2005	56 male	IIIa	Embolization	TV	Success	None
11	Poulios 2014	38 female	Ia/b	Surgery	NA	Success	None
12	Doppl 2007	36 male	Ia/b	Surgery	NA	Success	None
13	Gorospe 2012	59 male	Ia/b	Embolization failure with bailout surgery	TA	Success	None
14	Our case	70 male	Ia/b	Embolization	TA	Success	Severe mucus diarrhea after embolization
15	Athanasiou 2014	66 male	Ia/b	Embolization failure with bailout surgery	TA	Success	None
16	Noor 2016	61 male	Ia/b	Surgery	NA	Success	None
17	Metacalf 2008	50 male	Ia/b	Surgery	NA	Success	None
18	Hirota 2022	64 male	IIIa	Embolization failure with bailout surgery	TA	Success	AVM rupture after embolization
19	Cubisino 2021	72 male	Ia/b	No treatment	NA	NA	Improved during observation
20	Gupta 2019	46 male	Ia/b	Embolization failure with bailout surgery	TA	Success	None
21	Hendy 2018	24 male	Ia/b	Embolization	TA	Success	None
22	Brucher 2014	59 male	I	Embolization	TA	Success	None
23	Slutski 1988	63 male	I	Embolization	TA	Success	None
24	Okada 2002	69 female	I	Surgery	NA	Success	None
25	Kunioka 2019	57 male	IIIa	No treatment	NA	NA	Improved during observation
26	Hayakawa 1998	38 male	Ia/b	Surgery	NA	Success	None
27	Hajjar 2017	54 male	Ia/b	Embolization failure with bailout surgery	TA	Success	None
28	Kimura 2021	50 male	Ia/b	Surgery	NA	Success	None
29	Healy 2021	54 male	Ia/b	Embolization	TA	Success	None
30	Cheng 2017	62 male	IIIb	Surgery	NA	Success	None
31	Richard 2021	72 male	Ia/b	Embolization	TA	Success	None
32	Langroudi 2015	48 female	I	Surgery	NA	Success	None
33	Kai 2019	66 male	Ia/b	Presurgery embolization	NA	Success	None
34	Charalambous 2020	73 female	I	Embolization	TA	Success	None

AVM, arteriovenous malformation; NA, not available; TA, transarterial; TV, transvenous.

from 1988 to 2022 using the keywords “inferior mesenteric artery,” “arteriovenous malformation,” and “fistula” and found 39 cases. After excluding those without angiography [2], those with poor imaging quality [3], and those with an unclear relationship between the nidus and outflow veins in imaging examinations [4,5], we conducted an angioarchitecture analysis of 33 cases [6–40] in addition to our own (34 total cases), as listed in Table 1.

Three radiologists (C.Y., W.Y., and S.Y.) reviewed the definitions and typical angioarchitectures of each Yakes AVM type [1] before commencement of the angioarchitecture analysis. To more easily reach a consensus on AVM classification, the authors used the following rules for classification in addition

to the relevant lesion descriptions in the article when reviewing images of the enrolled cases:

1. Type I: No nidus can be identified. The feeder artery directly connects to the outflow vein/veins.
2. Type II: The nidus appears typical of a corkscrew, which precedes the outflow vein (IIb) or veins (IIa). A type II AVM can be identified by 3 distinct components: the feeder artery, the nidus, and the outflow vein.
3. Type III: The origin of the outflow vein is an aneurysmal dilatation, which appears as a venous sac. The nidus usually superimposes on the aneurysmal sac.

4. Type IV: The abnormal arteriovenous shunting/ microfistulas and the immediate outflow veins are small and arise from the colonic wall, indicating that the lesion primarily involves the capillary/microcirculation.

We compiled all reported cases and calculated the success rate of embolization and surgical resection respectively.

Case report

A 70-year-old man presented to the emergency department with lower abdominal pain, massive bloody diarrhea, and low blood pressure of 98/59 mmHg with tachycardia. Laboratory examinations showed a low hemoglobin level of 6.0 g/dL. Five days prior, he had experienced intermittent tarry stools, and endoscopy at another hospital revealed gastric varices with ulcers. His medical history included resection of a jejunal segment for ischemic enteritis 18 years previously, hyperlipidemia and diabetes mellitus under control by regular medication, and coronary arterial disease after stenting. He had no known history of viral or alcoholic hepatitis. In the emergency room, computed tomography (CT) angiography revealed swelling and poor enhancement of the sigmoid colon and rectum (Fig. 1A, arrows), which was suggestive of ischemic colitis. There was also a tangle of early enhanced corkscrew vessels (the nidus) in the pararectal fossa (Figs. 1A, asterisk and B, arrow) with an engorged mesocolic left marginal vein (Fig. 1B, arrowheads), raising suspicion for AVM. In addition, there was evidence of portal hypertension, including ascites, gastric varices, and obliteration of the main portal vein at the liver hilum with cavernous transformation (Fig. 2). Emergency colonoscopy revealed a bleeding ulcer at the rectosigmoid junction and edema of the colonic wall extending from the descending colon to the rectum, which was suggestive of congestive/ischemic colitis.

After discussing the case, the on-duty interventional radiologist, general surgeon, and gastroenterologist agreed to attempt embolization first because of the patient's age and unstable hemodynamic status.

The patient was sent to the angiographic suite. Selective inferior mesenteric arteriography confirmed an AVM comprising a nidus (Fig. 2, white arrow), a feeder from the superior rectal artery (Fig. 2, black arrows), and a single engorged and tortuous left mesocolic marginal vein (Fig. 2, arrowheads) draining to the portal cavernoma. According to the Yakes classification, the patient's AVM was classified as type IIb and its curative treatment is trans-arterial (TA) sclerosis of the nidus using ethanol or another liquid embolic [1].

After the superior rectal artery was catheterized, a mixture of N-butyl-2-cyanoacrylate with ethiodized oil (1:3-1:4) was injected into the nidus until it was completely devascularized. Postembolization angiography revealed no nidus or outflow vein (Fig. 3).

The first CT scan after embolization was performed 3 months later, revealing resolved ascites and a segment of edematous sigmoid colon, which may have caused the patient's postembolization mucus diarrhea. The nidus was completely glued with scattered glue casts were noted in the left meso-

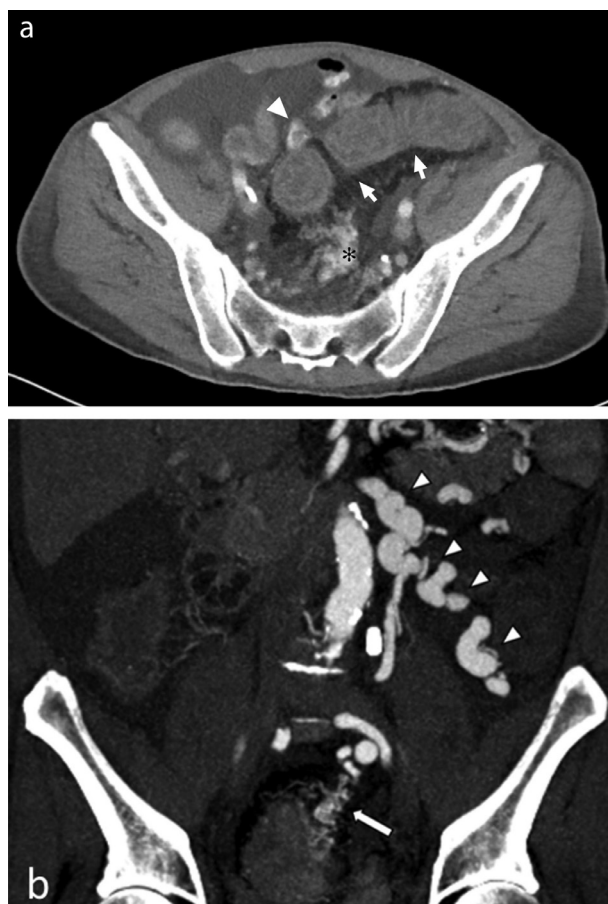


Fig. 1 – (A) Enhanced computed tomography revealed a poorly enhanced segment of the sigmoid colon (arrows). Note the well-enhanced small intestine (arrowhead) adjacent to the sigmoid colon. A cluster of abnormally engorged and tangled vessels, the nidus (asterisk), was noted in the pararectal fossa. (B) Enhanced computed tomography revealed a cluster of abnormally engorged and tangled vessels, the nidus (arrow), in the pararectal fossa. A segment of engorged left mesocolic marginal vein was noted (arrowheads).

colic marginal vein. The patient was healthy and symptom-free 2 years after embolization, and a second CT scan was performed. The edematous sigmoid colon had returned to normal, and the glue casts in the nidus and the left mesocolic marginal vein had disappeared. No ascites was present, but the portal cavernoma and gastric varices remained.

Discussion

During the 14-year period of our literature review, 39 case reports were published; 34 involved men and 5 involved women. The patients' mean age was 57.4 years (range, 24-76 years). Among 27 patients without a history of surgery or trauma, 26 were male and only 1 was female. Among 23 patients with ischemic colitis (23/39 [58.9%]), 14 (60.9%) developed lower gastrointestinal tract bleeding.

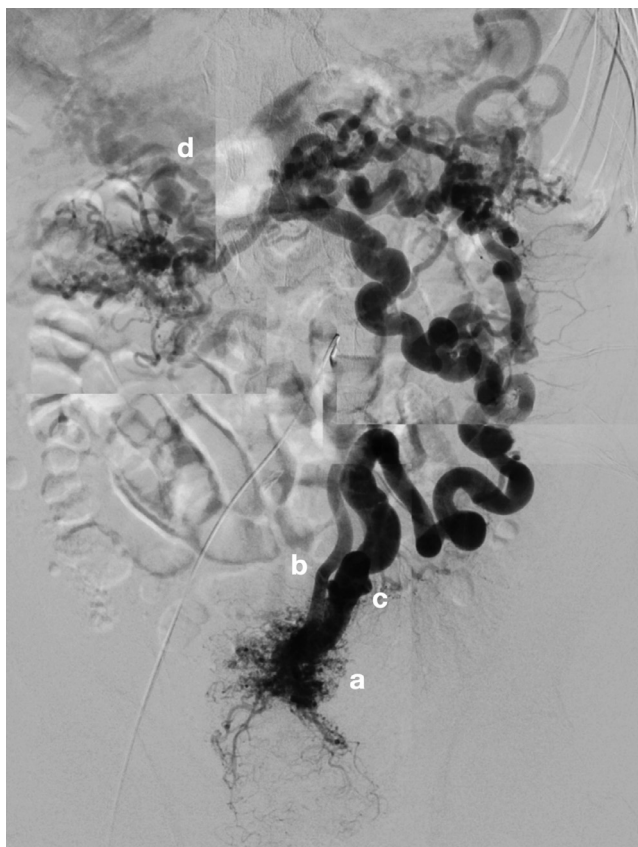


Fig. 2 – Pre-embolization angiography of type IIb arteriovenous malformation. (A) The nidus was fed by (B) the superior rectal artery and drained by (C) a single engorged left mesocolic marginal vein. (D) Note the portal cavernoma at the liver hilum.

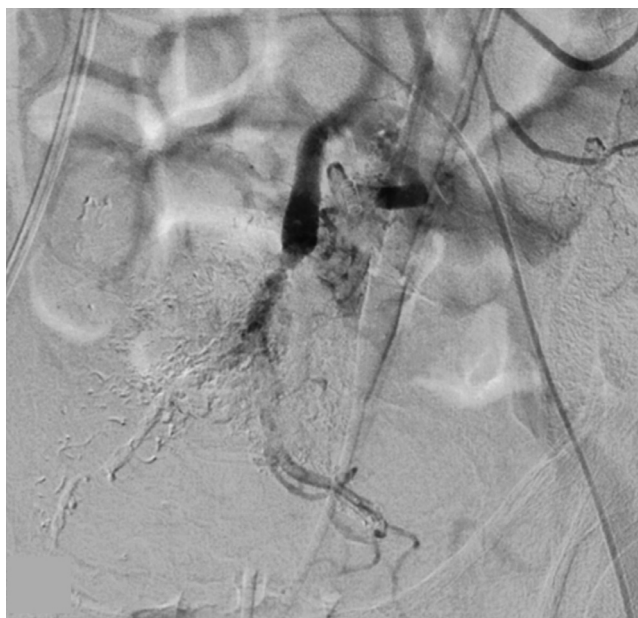


Fig. 3 – Postembolization arteriography showed no visible nidus and the outflow vein.

Table 2 – The Yakes type-specific success rates for embolization and surgery.

Yakes classification	Case count	Embolization	Surgery
Type I	5	100% (4/4)	100% (2/2)
Type IIa/b	21	58.3% (7/12)	100% (9/9)
Type IIIa	5	100% (2/2)	100% (3/3)
Type IIIb	2	NA	100% (2/2)
Type IV	2	NA	100% (2/2)

Angioarchitecture analysis

The locations and outflow veins of the reported AVMs and AVFs are illustrated in Fig. 4. The feeder artery of the nidus was the IMA and its branches. The outflow veins of the nidus/fistula included the superior rectal vein, the inferior mesenteric vein, the left mesocolic marginal vein, and veins in the colonic wall.

In total, 43 AVMs/AVFs in 35 patients were enrolled for angioarchitecture analysis. If a patient had 2 AVMs, such as Patient 12, the AVMs were labeled 12-1 and 12-2, respectively. For a type IV AVM, even if the microfistulas extended from the sigmoid to the descending colon, they were considered as one AVM. According to the Yakes classification, there were 27 (62.8%) type IIa/b AVMs, 6 type IIIa AVMs, 6 type I AVMs, 2 type IIIb AVMs, and 2 type IV AVMs (Fig. 5).

There were 22 cases of embolization, 2 of which were presurgical embolization to avoid bleeding during surgery [25,35]. Among all cases of curative-intent treatment, there were 20 embolizations, 6 of which failed (symptoms either did not improve or worsened) [7,19–21,29,33]. Thus, the embolization success rate was 70.0% (14/20). The Yakes type-specific success rates are listed in Table 2.

The success rate of all 23 surgeries in 23 patients was 100%, including 16 primary surgeries and 7 bailout surgeries (1 after a portal vein stenting without symptoms improvement and 6 surgeries after failed embolization). Six cases of embolization failure via the TA route were due to persistent or recurring symptoms in the short term [7,12,29], deterioration of ischemia [19,20], technical issues [21], and AVM rupture after embolization [33]. Among the 9 cases of successful embolization via the TA route, our patient had frequent diarrhea, which disappeared after 6 months; 1 patient had paroxysmal abdominal pain after embolization [16], which disappeared after a few days; and the remaining 7 patients had no complications after embolization and recovered well [11,13,27,32,34,35,40]. All 5 patients who underwent successful transvenous (TV) route embolization [10,14,26,28,31] developed no complications and recovered well (Fig. 6).

The curative intervention for Yakes type II AVM/AVF is complete sclerosis of the nidus using a liquid embolic such as ethanol [32], glue (our case), foam [10], or onyx [11,13,14,20]. However, the use of a liquid embolic requires experience; incomplete devascularization of the nidus can lead to persistent symptoms [29] or symptom recurrence [7]. For peripheral AVMs, there are 3 ways to deliver a liquid embolic to the nidus: direct puncture of the nidus, direct puncture of the

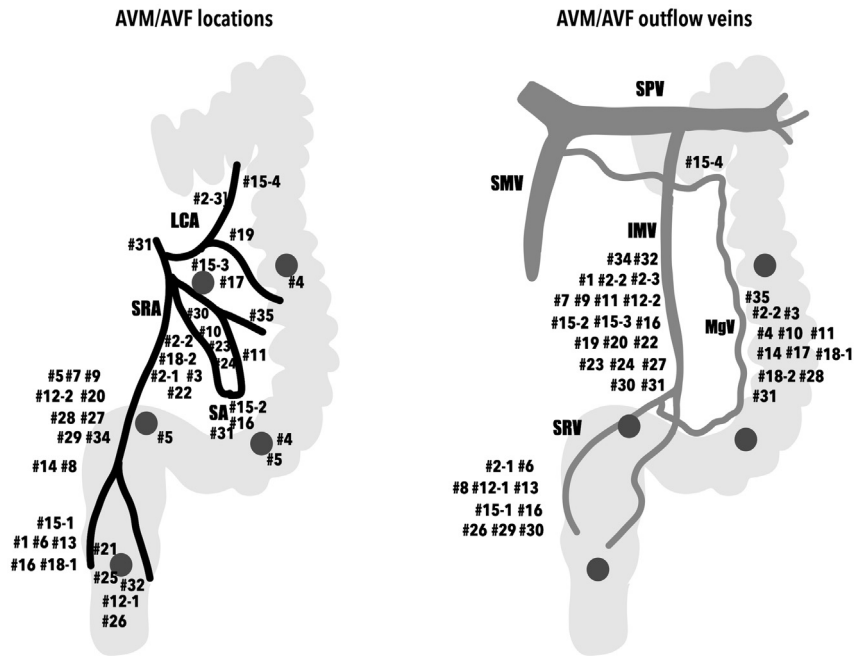


Fig. 4 – Locations and outflow veins of the reported arteriovenous malformations and arteriovenous fistulas. The authors created the illustration only for this case report. IMV, inferior mesenteric vein; LCA, left colic artery; MgV, marginal vein; SA, sigmoid artery; SMV, superior mesenteric vein; SPV, splenic vein; SRA, superior rectal artery; SRV, superior rectal vein.

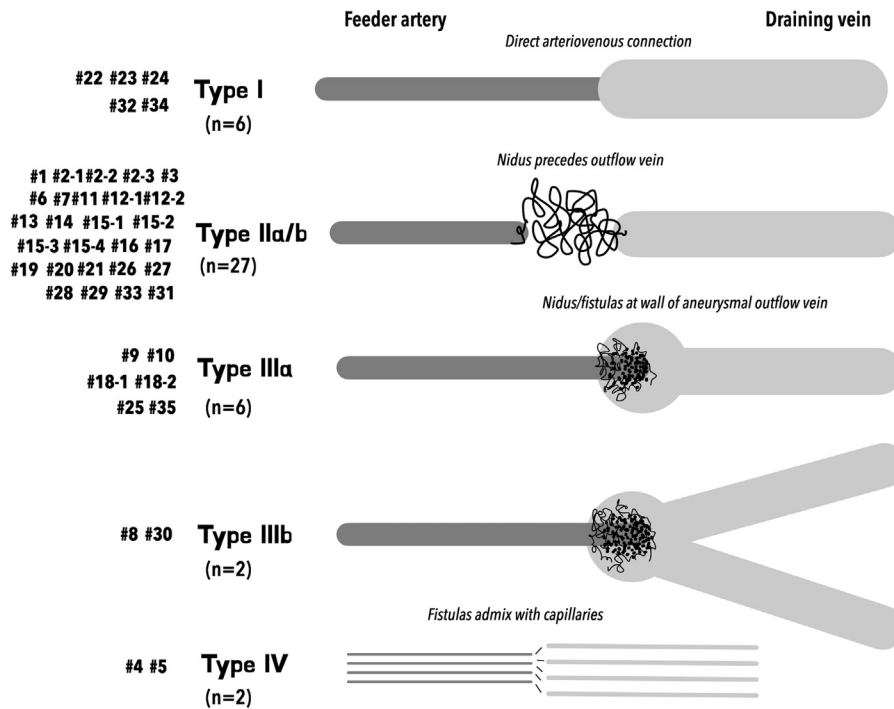


Fig. 5 – Results of angioarchitecture analysis of arteriovenous malformations and arteriovenous fistulas according to Yakes types. The authors created the illustration only for this case report.

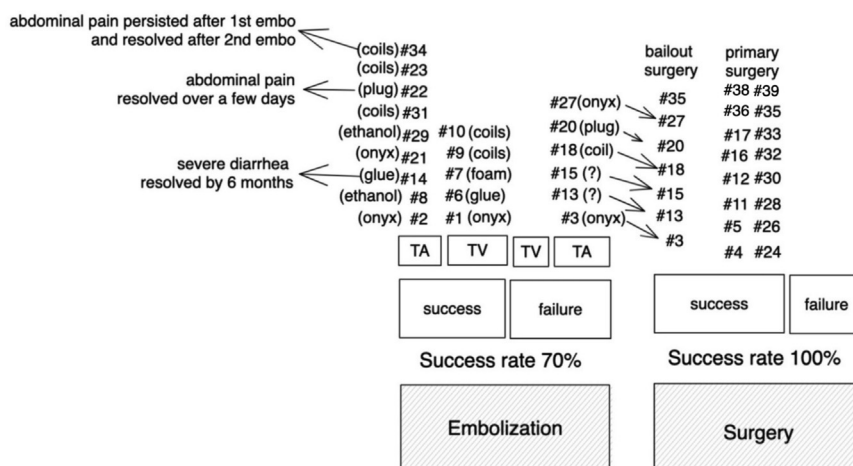


Fig. 6 – Results of literature review on treatment outcomes. The authors created the illustration only for this case report. TA, transarterial; TV, transvenous.

feeder arteries to the nidus, and catheterization of the feeder arteries. For visceral AVMs, direct puncture of the feeder artery or the nidus for injection of the sclerosing agent is dangerous and usually impossible. Catheterization of the feeder artery is the best approach in such cases. For Yakes type III AVM/AVF, interventionists can choose to completely sclerosed the nidus or completely occlude the outflow vein with coils [26,28], plugs [29], or a liquid embolic [10,14,31]. Because a liquid embolic is not easy to control, the TV route should be attempted first using nonliquid embolics such as coils or plugs, with which general interventionists are familiar. Catheterization of the outflow vein for embolization (TV route) is the safest technique, with no reported procedure-related or postembolization complications. However, if main portal vein thrombosis is present or the nidus outflow vein is too tortuous to traverse, direct puncture of the aneurysmal venous sac for embolization is an alternative option [29].

For our patient, transhepatic access to the left mesocolic marginal vein was impossible because of main portal vein obliteration with cavernous transformation. In addition, direct puncture of the left mesocolic marginal vein posed a risk of bowel perforation, so we chose the TA route. For a type IIb AVM, TA nidus sclerosis is the recommended curative treatment. However, the major concern of the TA route for delivery of a liquid embolic is the risk of inadvertently occluding the vasa recta, which may worsen ischemic colitis [20,33].

Should the multidisciplinary team choose surgery or embolization to treat an IMA AVM/AVF? From the patient’s perspective, because the success rate of surgical treatment is 100% and no significant complications have been reported, surgery should be the first choice if the patient’s vital signs are acceptable, and the surgeon and anesthesiologist consider that such treatment is feasible. However, if the patient is unsuitable for general anesthesia and surgery, such as our patient (whose bloody diarrhea caused hemodynamic instability), then endovascular embolization should be considered first.

Because Yakes type I AVF is easy and safe to treat percutaneously, surgery should not be the first choice. Yakes type IV

AVM/AVF is challenging to cure percutaneously, and surgery should be considered first. Most reported cases of AVMs/AVFs are Yakes type IIa/b; therefore, TA nidus sclerosis using a liquid embolic should be the first choice. For type III AVM, the TV route for embolization should be considered first because it has a 100% success rate without reported postembolization complications.

Data availability statement

Data may be obtained from the corresponding author.

Past presentation

None.

Patient consent

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

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