



Case report

A case of arteriovenous malformation in the inferior mesenteric artery region resected surgically under intraoperative indocyanine green fluorescence imaging

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ABSTRACT

Introduction and importance: An arteriovenous malformation (AVM) is defined as a vascular malformation with a short, non-capillary communication between the arteries and veins. Most gastrointestinal AVMs are solitary, occurring predominantly in the stomach, small intestine and right colon, and rarely in the inferior mesenteric artery (IMA) region.

Case presentation: A 70-year-old man was first diagnosed with ischemic enteritis two years earlier, and was hospitalized several times with the same diagnosis. He visited our hospital because of left lower abdominal pain and melena. Colonoscopy showed findings suggestive of ischemic enteritis, and contrast-enhanced computed tomography (CT) and IMA angiography showed hyperplasia and dilation of blood vessels from the sigmoid-descending colon junction to the upper rectum. We performed conventional laparoscopic low anterior resection using intraoperative intravenous injection of indocyanine green (ICG). The final diagnosis was arteriovenous malformation in the IMA region. The patient had an uneventful postoperative course and was discharged on the 13th day after the operation.

Clinical discussion: Cases of AVM in the IMA region are relatively rare. This is the first reported case of AVM in the IMA region that was resected under intraoperative ICG fluorescence imaging (FI), which provided useful information on the extent of intestinal resection and mesenteric dissection required, and confirmed the adequacy of intestinal blood flow during and after mesenteric dissection and anastomosis.

Conclusion: It is advisable to use ICG FI intraoperatively during resection of AVMs in the IMA region, as with colorectal cancer surgery.

1. Introduction

An arteriovenous malformation (AVM) is defined as a vascular malformation with a short, non-capillary communication between the arteries and veins [1]. Gastrointestinal AVMs occur most often in the stomach, small intestine and right colon, and only rarely in the inferior mesenteric artery (IMA) region [2]. Colonoscopy, computed tomography (CT) and angiography are all useful for the diagnosis of gastrointestinal AVMs. Recently, indocyanine green fluorescence imaging (ICG FI) has become widespread in several medical and surgical specialties,

including in colorectal surgery [3–5]. We report here an extremely rare case of an AVM in the IMA region that was surgically resected under intraoperative ICG FI. This report has been reported in line with SCARE criteria [6].

2. Case presentation

A 70-year-old man under medical treatment for hypertension and dyslipidemia visited our hospital because of left lower abdominal pain and melena. He had no past history of surgery, although he had been

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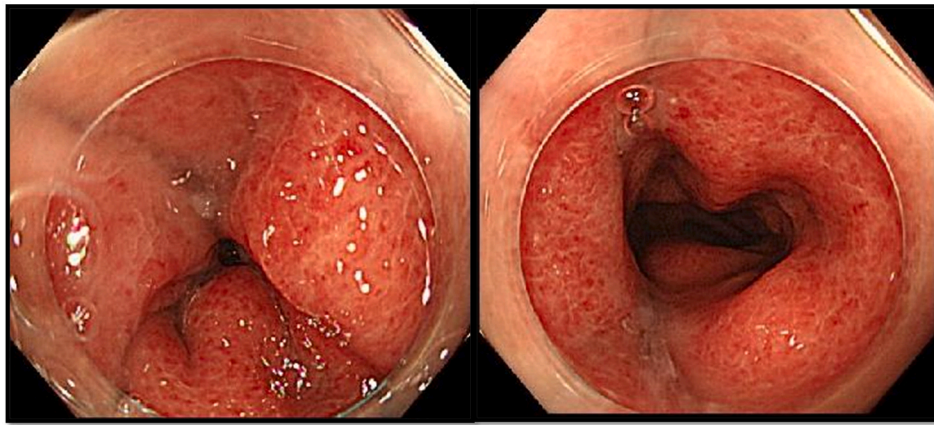


Fig. 1. Colonoscopy findings.

Ischemic enteritis and stenosis were observed from the sigmoid-descending colon junction to the upper rectum.

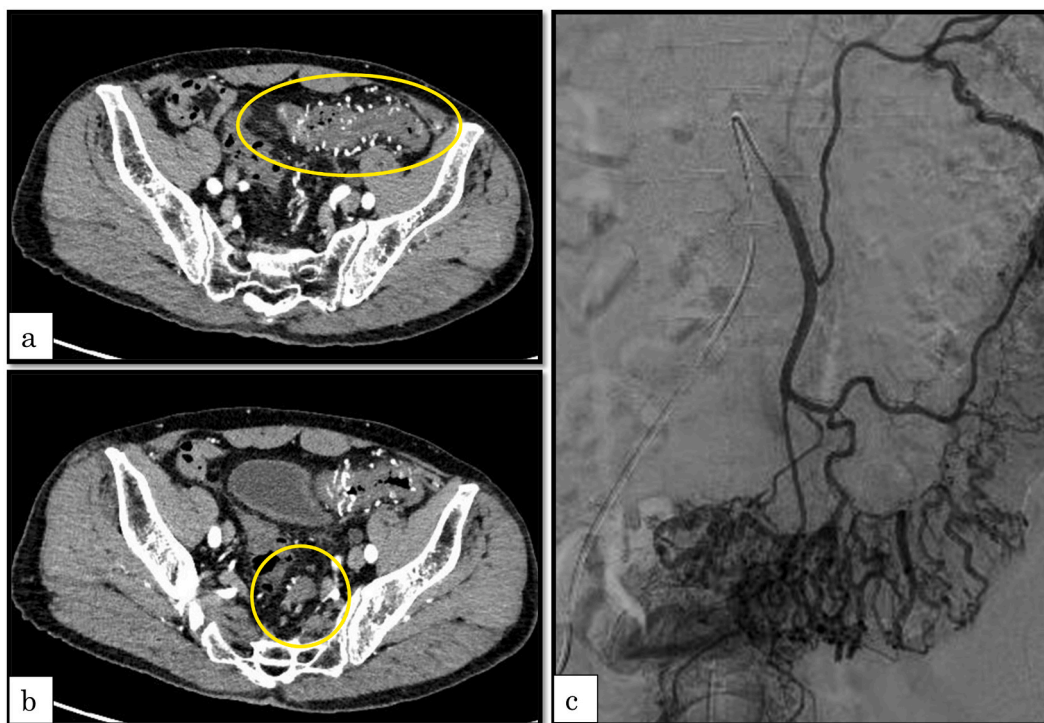


Fig. 2. Contrast-enhanced computed tomography and inferior mesenteric artery (IMA) angiography findings.

a) Extensive intestinal wall thickening, hyperplasia and dilation of blood vessels near the intestinal tract were seen in the IMA region

b) The caudal-most part of the nidus was at the level of the peritoneal reflection on the left side of the rectum

c) Hyperplasia and dilation of blood vessels from the sigmoid-descending colon junction to the upper rectum were present.

diagnosed with ischemic enteritis two years earlier, and had been hospitalized several times with the same diagnosis. Colonoscopy on admission showed findings suggestive of ischemic enteritis, with mucosal edema, redness and stenosis from the sigmoid-descending colon junction to the upper rectum (Fig. 1). Several biopsies from the lesion revealed no evidence of malignancy. Contrast-enhanced CT showed extensive intestinal wall thickening, hyperplasia and dilatation of blood vessels near the intestinal tract in the IMA region, and the caudal-most nidus was at the level of the peritoneal reflection on the left side of the rectum (Fig. 2a, b). IMA angiography also showed hyperplasia and dilatation of blood vessels from the sigmoid-descending colon junction to the upper rectum (Fig. 2c). No abnormal blood vessels were found on superior mesenteric artery angiography. Based on a diagnosis

of AVM in the IMA region, we decided to perform surgery for resection of the AVM.

We performed conventional laparoscopic low anterior resection to completely resect the AVM. Laparoscopic observation revealed findings of intestinal wall and mesenteric thickening from the sigmoid-descending colon junction to the upper rectum, and hyperplasia of blood vessels around the lesion (Fig. 3a). We performed ICG FI to confirm intestinal and mesenteric blood flow (Fig. 3b). After mobilization of the resected intestinal specimen along with the AVM, a 7 cm long midline laparotomy incision was made, intestinal and mesenteric blood flow was confirmed again by ICG FI after mesenteric dissection, and the specimen was removed from the abdomen (Fig. 3c). We performed reconstruction with a double stapling technique anastomosis and

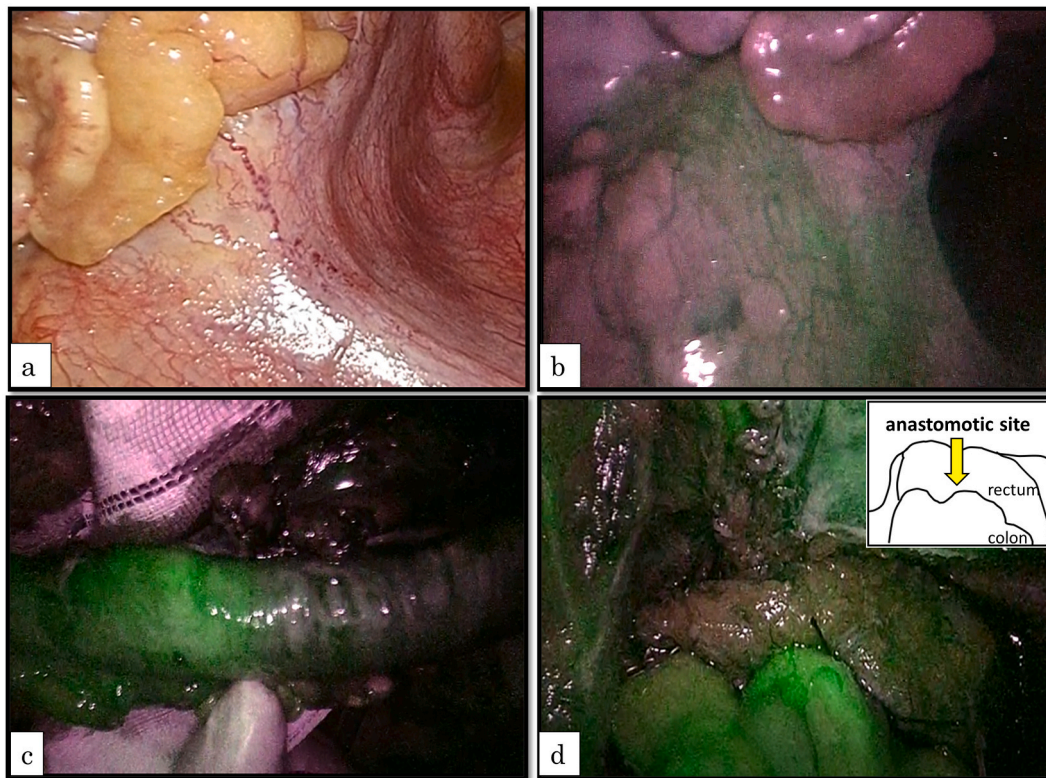


Fig. 3. Operative findings.

- Intestinal wall and mesenteric thickening in the inferior mesenteric artery (IMA) region
- Indocyanine green (ICG) fluorescence imaging (FI) showed diminished fluorescence of the intestinal wall and mesenteric thickening, along with enhanced fluorescence in the surrounding area of hyperplasia.
- We confirmed the adequacy of intestinal and mesenteric blood flow by ICG FI after mesenteric dissection
- We confirmed the adequacy of blood flow at the anastomotic site by ICG FI.

confirmed blood flow at the anastomotic site using ICG FI (Fig. 3d). The surgical duration was 327 min, and total blood loss was 121 g.

Evaluation of the excised specimen showed that the intestinal tract was edematous, thickened and narrowed, and the mesentery was swollen and stiff (Fig. 4). Histopathological examination revealed many large and small blood vessels growing in a nodular manner, along with dilatation of the blood vessels and irregularly distributed blood clots. The blood vessels were randomly thickened, thinned, and lacked elastic fibers in the blood vessel wall, and, for many of them, it was unclear whether they were arteries or veins (Fig. 5). The final diagnosis was AVM in the IMA region.

The patient had an uneventful postoperative course and was discharged on the 13th postoperative day.

3. Discussion

Since Margulis et al. first reported on gastrointestinal AVMs in 1960, the number of reported cases of AVM has increased due to the widespread use of colonoscopy and increase in the number of elderly people with arteriosclerosis [7]. Its causes are broadly divided into congenital, due to abnormal vascular development during the embryonic period, and acquired, due to trauma, surgery, pregnancy and delivery [1]. Most gastrointestinal AVMs are solitary, occurring predominantly in the stomach, small intestine and right colon, and rarely in the IMA region [2]. Gastrointestinal AVMs are often discovered during evaluation of gastrointestinal bleeding, and are widely recognized as one of the potential causes of lower gastrointestinal bleeding [8]. In our patient, since there was no obvious cause of the AVM, it was considered an idiopathic AVM in the IMA region. In addition, his main complaint of left lower abdominal pain during defecation was considered as being not only due

to ischemic enteritis, but also intestinal obstruction due to relatively extensive stenosis because of the AVM.

The colonoscopy findings of gastrointestinal AVM are typically suggestive of ischemic enteritis, with multiple ulcers and mucosal redness, hyperemia and edema [9]. Ischemic colitis is generally considered to be more likely to occur in patients with hypertension and diabetes, because vascular factors such as arteriosclerosis and vasospasm, and intestinal factors such as increased peristalsis, cause ischemia of the intestinal wall. In addition, the AVMs cause ischemic enteritis by the clinical phenomenon of the steal syndrome, in which arterial blood flows directly into the veins in the vascular nidus, reducing organ blood flow. Additionally, the subsequent secondary increases in venous pressure cause marked congestion in the organs, further exacerbating ischemia [10]. CT is useful for ruling out malignancy in cases with intestinal wall thickening, and identifying the source of bleeding in the presence of melena, but not for direct diagnosis of AVMs. On the other hand, abdominal angiography is important for diagnosis of the AVM, which is identified by the characteristic findings of abnormal enhancement of the intestinal wall in the arterial phase, early enhancement of the outflow vein, and enhancement of the outflow vein that is prolonged to the late venous phase [11]. Our case was consistent with AVM based on the characteristic colonoscopy and abdominal angiography findings.

The treatment methods of AVM include interventional radiology (IVR), endoscopic treatment and surgical treatment, although there are no clear criteria for treatment selection. The advantages of IVR and endoscopic treatment are that they can be performed repeatedly, are relatively minimally invasive, have a low cost, and can be performed in patients with a high risk during surgery or without subjective symptoms, such as melena and abdominal pain. However, these procedures might



Fig. 4. Macroscopic evaluation of the excised specimen. The intestinal tract was edematous, thickened and narrowed, and the mesentery was swollen and stiff.

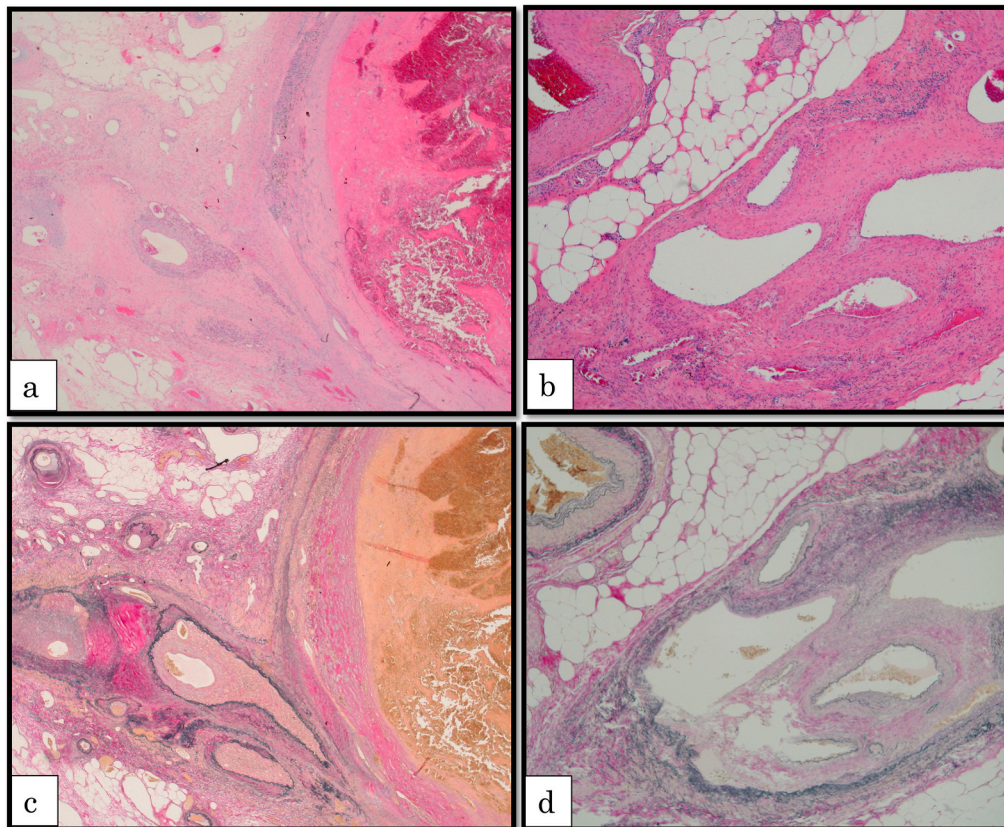


Fig. 5. Histopathological evaluation of the excised specimen. a, b) Immunostaining (Hematoxylin-Eosin): Many large and small blood vessels grew in a nodular manner, along with dilatation of the blood vessels and irregularly distributed blood clots. c, d) Immunostaining (Elastica van Gieson): The blood vessels were variously thickened, thinned, and lacked elastic fibers in the blood vessel wall, and, for many of the vessels, it was unclear whether they were arteries or veins.

also require transition to surgery due to AVM recurrence or an effect of the treatment. Hence, surgical resection is usually considered as the first-line treatment. In our case, we chose surgical treatment for early improvement of symptoms, because the patient was forced to eat an easily digestible meal, the frequency of recurrent attacks of abdominal pain was increasing, and the pain was gradually increasing in severity. The selected surgical procedure should be one that allows intestinal resection, including that of the AVM, while minimizing blood flow to the AVM. We opted for a laparoscopic low anterior resection in our case because of the presence of abnormalities in the intestinal tract, mesentery and blood vessels in a wide part of the IMA area, and to ensure excision of the caudal-most part of the nidus and the entire abnormal intestinal tract and mesentery recognized by preoperative imaging and intraoperative findings. When the abnormality caused by the AVM is long-standing and widespread, as in our case, it is considered difficult to improve the intestinal condition and symptoms by any treatment method other than surgery, due to the presence of irreversible changes in the mesenteric blood vessels. Therefore, our case was a good candidate for surgery.

It is difficult to accurately determine the range of abnormalities caused by AVM. Preoperative CT and angiography can predict the range of abnormalities in the blood vessels and intestinal tract caused by the AVM to some extent, and intraoperative colonoscopy is useful for determining the appropriate extent of intestinal resection that would allow maximum intestinal preservation. On the other hand, ICG FI has become widespread in several medical and surgical specialties, such as for determining cardiac output and hepatic function and performing ophthalmic angiography [3,4]. Previous reports showed that ICG FI contributed to reducing the risk of postoperative anastomotic leakage, and that ICG injection into the submucosal layer enables intraoperative visualization of lymphatic flow and can be used for intraoperative navigation in colorectal cancer surgery [5,12,13]. We performed a PubMed search using keywords such as “ICG” and “arteriovenous malformations”, and found that this is the first reported case of an AVM in the colorectal region that was resected under ICG FI, although some previous reports have shown the utility of ICG FI for AVMs in the small intestinal region [14–16]. In our case, the distribution of abnormal blood vessels caused by the AVM was consistent with the thickened mesentery, weak ICG fluorescence of the intestinal tract and mesentery in this region was suggestive of ischemic enteritis, and the areas of enhanced ICG fluorescence were due to angiogenesis around the region. In addition, because there was abundant hyperplasia around the AVM and the extent to which the AVM contributed to intestinal blood flow was not known, ICG FI was useful and important for confirming the adequacy of intestinal blood flow after mesenteric dissection.

4. Conclusion

As with colorectal cancer surgery, ICG FI during surgery for AVMs in the IMA region provides useful information about the extent of intestinal resection and mesenteric dissection required, and the intestinal blood flow at the site of anastomosis.

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Ethical approval

Not applicable.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Registration of research studies

1. Name of the registry:

A case of arteriovenous malformation in the inferior mesenteric artery region resected surgically under intraoperative indocyanine green fluorescence imaging guidance

2. Unique identifying number or registration ID: researchregistry7461

3. Hyperlink to your specific registration (must be publicly accessible and will be checked): <https://www.researchregistry.com/register-now#home/>

Guarantor

Toshio Shiraishi.

CRediT authorship contribution statement

Toshio Shiraishi was responsible for the study concept. All authors have read and approved the final version of the manuscript.

Declaration of competing interest

All authors declare that have no conflict of interest.

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