

Gastrointestinal Bleeding in Small Bowel Arteriovenous Malformation: A Case Report

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ABSTRACT

Arteriovenous malformations of the gastrointestinal tract (AVM) can occasionally result in gastrointestinal bleeding. Less than 5% of nonvariceal upper gastrointestinal bleeding is caused by gastrointestinal AVMs, a relatively uncommon cause of GI bleeding. Clinical symptoms that they might present include intussusception, persistent anemia, overt or obscure GI bleeding, and abdominal pain. A 41-year-old male patient presented with a 7-year history of recurrent melena. An initial enteroscopy showed spurting bleeding in the jejunum with histological examination showing AVM with ectopic pancreas. The patient underwent laparotomy with resection, anastomosis, and the construction of a new duodenojejunosomy. The patient complained of recurrent melena during three years of postoperative follow-up. The patient underwent laparotomy resection of the ileum to ascending colon, ileostomy, and splenectomy. Histological examination of the bleeding duodenal mucosa confirmed an arteriovenous malformation. The patient then underwent endoscopic hemostasis using hemoclips and was treated with coil embolization. In abdominal CT, angiographic evaluation showed no visible picture of vascular malformations. Intestinal AVMs are rare but should be considered as the differential diagnosis in patients with recurrent gastrointestinal bleeding. Endoscopic hemostasis, surgical resection, and angiographic intervention can be considered as treatments for AVM.

Keywords: Arteriovenous malformation, gastrointestinal bleeding, small bowel

ABSTRAK

Malformasi Arteri-Vena (MVA) adalah salah satu penyebab langka dari perdarahan saluran cerna bagian atas. Kurang dari 5% penyebab perdarahan gastrointestinal nonvariseal disebabkan oleh MVA. Manifestasi klinis MVA dapat berupa intususpsi, anemia persisten, perdarahan gastrointestinal yang jelas atau samar, dan nyeri abdomen. Seorang laki-laki berusia 41 tahun datang dengan riwayat melena rekuren selama 7 tahun. Enteroskopi awal menunjukkan perdarahan menyembur di jejunum dan pemeriksaan histopatologi menunjukkan MVA dengan pankreas ektopik. Pasien selanjutnya menjalani laparotomi dengan reseksi, anastomosis, dan pembentukan duodenojejunosomi. Setelahnya, pasien masih mengalami melena rekuren selama tiga tahun follow-up pasca operasi. Pasien lalu menjalani laparotomi reseksi dari ileum hingga kolon ascendens, ileostomi, dan splenektomi. Pemeriksaan patologi dari mukosa duodenum yang berdarah menunjukkan MVA. Pasien lalu dilakukan hemostasis endoskopik menggunakan hemoklip dan embolisasi koil. Pada CT abdomen, evaluasi angiografik menunjukkan resolusi dari MVA. Meskipun langka, MVA intestinal harus dipertimbangkan sebagai diagnosis diferensial pada kasus perdarahan gastrointestinal rekuren. Hemostasis endoskopik, reseksi surgical, dan intervensi angiografik dapat dilakukan untuk menatalaksana MVA.

Kata kunci: Malformasi arteri-vena, pendarahan gastrointestinal, usus halus

INTRODUCTION

Arteriovenous malformations of the gastrointestinal tract (AVM) can occasionally result in gastrointestinal bleeding. Less than 5% of nonvariceal upper gastrointestinal bleeding is caused by gastrointestinal AVMs, a relatively uncommon cause of GI bleeding. Clinical symptoms that they might present include intussusception, persistent anemia, overt or obscure GI bleeding, and abdominal pain.² In patients over 50 years old, AVMs are the leading cause of bleeding. AVMs are linked to various health issues, such as chronic renal disease and valvular heart disease. AVMs are more prevalent in older adults. Endoscopy, enteroscopy, x-ray studies, capsule endoscopy, deep small bowel enteroscopy, and intraoperative enteroscopy are some of the several tests that may be performed to identify and treat the cause of small intestine hemorrhage. Typically, cautery via an endoscopy or enteroscopy is used to treat AVMs.³

Because of their position, small intestinal AVMs can be difficult to detect, even if it is actively bleeding. Endoscopic therapy and interventional radiology are the primary therapeutic techniques for bleeding small intestinal vascular lesions. Although coil embolization of the abnormality is safe, there is always the possibility of ischemia of that specific bowel segment. Surgery is the final option for severe small intestinal hemorrhage if endoscopic and radiographic treatments have failed.⁴

CASE ILLUSTRATION

A 41-year-old male patient presented with a 7-year history of recurrent melena. He had no history of bleeding disorder and was not on anticoagulant therapy. History of bloody vomiting or nausea was denied. In the past four years ago, he had known arteriovenous malformations (AVMs) duodenum. On initial physical examination, the patient was revealed to be moderately ill, fully alert, with typical vital signs, pale conjunctiva, and without epigastric pain. His laboratory test findings showed normocytic anemia; the other values were within the normal range.

An initial enteroscopy showed spurting bleeding in the jejunum, with histological of the jejunum showing arteriovenous malformation (AVM) with an ectopic pancreas (Fig. 1A). The patient underwent laparotomy with resection, anastomosis, and construction of a new duodenojejunostomy. The patient complained of recurrent melena during three years of postoperative follow-up. The patient underwent esophagogastroduodenoscopy and colonoscopy,

which revealed multiple ulcers in the duodenal and jejunum. Enteroscopy revealed small bowel bleeding and ileocolitis. The abdominal CT angiography showed dilatation of the superior mesenteric artery and vein to the duodenum, suggesting the presence of AVM. The patient underwent laparotomy and resection of the ileum to ascending colon, ileostomy, and splenectomy. Histological examination of the bleeding duodenum mucosa confirmed an arteriovenous malformation.

There was recurrent melena and anemia that persisted seven months after laparotomy resection of the ileum to the ascending colon. The patient underwent endoscopic hemostasis using hemoclips and was treated by coil embolization (Fig. 1B). A subsequent abdominal CT angiography revealed no visible picture of duodenal vascular malformation that gets feeding from the second branch superior mesenteric artery and possibly also get feeding from pancreaticoduodenalis anterior and posterior artery (Fig. 2A and B).

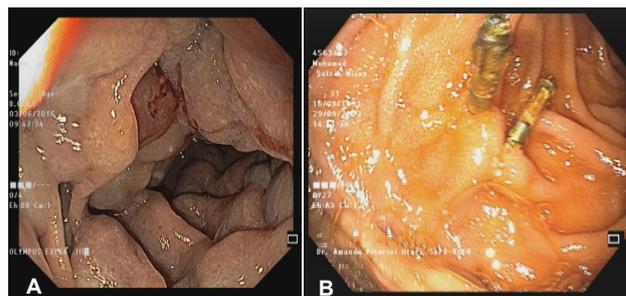


Fig. 1 A) Enteroscopy view showed spurting bleeding in the jejunum B) Endoscopic hemostasis using hemoclips



Fig. 2 A) and B) Abdominal CT angiography evaluation

DISCUSSION

Arteriovenous malformations (AVMs) are direct connections between arteries and veins without the presence of a capillary bed from an embryonic failure in the vascular development of the affected regions. Histopathology of AVM is identified as aberrant vessels with thickened, hypertrophic walls that vary widely in thickness. AVM is a chronic condition of

dilatation of submucosal and mucosal vessels. AVM might present as part of a congenital disorder, such as Henoch Schonlein Purpura, Hereditary Hemorrhagic Telangiectasia (HHT), and Ehlers-Danlos.^{3,5} AVM in the small bowel causes occult gastrointestinal bleeding or persisting gastrointestinal bleeding.⁶ The authors describe the case of a man, 41-year-old with recurrent gastrointestinal bleeding with multiple AVM in small bowels, including duodenum and jejunum, and it is a rare case. AVM is prone to rebleeding. Thus, surgical resection is usually needed.

AVM is one cause of small bowel bleeding. Patients with small bowel bleeding can present with overt, occult, or obscure GI bleeding. Clinicians must first assess the clinical stability in patients with presumed small bowel bleeding. If hemodynamically stable, then patients can be managed in an outpatient setting. The initial diagnostic modalities are upper endoscopy and colonoscopy. A repeat or extensive endoscopy is usually performed if the initial examination is negative. If still negative, Video Capsule Endoscopy (VCE) or multiphasic CT enterography can be performed. Once the bleeding source is identified, clinicians can perform push or balloon-assisted enteroscopy as diagnostic and therapeutic modalities. For clinically unstable patients with brisk bleeding, then CT angiography would be preferred.⁷

It is still challenging to identify and properly diagnose GI tract vascular malformations. AVMs can be asymptomatic and discovered incidentally through endoscopy. It can also cause painless gastrointestinal bleeding and chronic anemia.^{8,9} In 2005, a strategy for detecting and treating obscure GI bleeding was proposed. EGD and colonoscopy should be done on individuals with obscure GI bleeding. Overall, 20-30% of patients assessed for obscure bleeding have upper and lower GI tract lesions that may be dismissed during prior endoscopic evaluation.¹⁰

According to a recent meta-analysis, the diagnosis of current bleeding by CT angiography has a sensitivity of 89% and a specificity of 85%.¹¹ Additionally, video capsule endoscopy or double-balloon endoscopy might be effective in identifying AVMs in the small intestine.¹² Lesions such as erosions, ulcers, vascular lesions, cancers, and others that cause bleeding in the small intestine can be identified by capsule endoscopy.¹³

Intestinal AVMs may be categorized into three groups based on Moore's classification. Type 1 AVMs are an acquired condition that usually affects older people and manifests in the right colon. Type 2 AVMs generally display in the small intestine in younger

people and are thought to represent a congenital condition. Type 3 AVMs manifest in individuals with inherited hemorrhagic telangiectasia as GI involvement. Type 1 AVMs, thought to be acquired diseases, are more frequently seen in the right colon, where intestinal tension is high.^{8,14,15}

It is uncommon for gastrointestinal AVMs to bleed. About 5% of nonvariceal upper gastrointestinal bleeding is caused by angiodysplasias or vascular abnormalities. AVM lesions in the gastrointestinal system can cause symptoms such as hematemesis, melena, or iron deficiency anemia. When determining the source of bleeding during esophagogastroduodenoscopy, the existence of AVM should be suspected. Endoscopy is the most often used approach in treating these patients, with CT angiography reserved for situations with challenging diagnostic clarification.¹⁶

Large duodenal AVMs are uncommon, and prior case reports have documented successful therapy of bleeding lesions with angiographic embolization. This method is not universally effective, nor has laparoscopic closure of the gastroduodenal artery feeding branch.¹⁷ Abnormal blood vessels, such as angioectasias, AVMs, hemangiomas, and Dieulafoy's lesions, cause 20 to 30% of small intestinal hemorrhage. AVMs are the most prevalent cause of small-bowel hemorrhage in adults over the age of 50.^{9,18}

The choice of evaluation and treatment of AVM depends on the epidemiology, pathology, and clinical presentation. Most small intestinal AVMs need to be surgically removed due to their size and tendency to rebleeding. Finding the lesion during surgery might be challenging in patients who require surgical procedures for small intestinal AVMs. According to reports, mesenteric angiography and subsequent microcoils embolization are efficient for primary hemostasis and preoperative localization. Additionally, intraoperative indocyanine green injections and endoscopic tattooing or marking clips are allegedly helpful for localizing the target lesion.¹⁴ Microcoils can also be applied as a radiographic marker during surgery to point out the location of the bleeding source.¹⁰

Minimally invasive therapy was made possible by preoperative identification of the small intestinal lesion.¹⁹ Previous research found that older versions of the endoclip were less effective at achieving hemostasis if the bleeding artery was larger than two mm.²⁰ Surgery is frequently necessary when an AVM is bleeding. A case of stomach AVM effectively treated by endoclip application and percutaneous transarterial coil embolization is described by McCrary et al.¹

Around 20% of patients with lower GI hemorrhage may experience early recurrent bleeding despite the reported high 96% success rate of rapid hemostasis obtained by embolization. The reported incidence of serious sequelae, such as artery dissection and intestinal infarction, was 17%, which is a significant restriction of angiographic embolization.^{21,22}

In this case, the patient had multiple surgical resections but still had recurrent melena. The patient also performed endoscopic marking clips and was treated by coil embolization. In abdominal CT, angiography evaluation showed no visible picture of vascular malformations.

CONCLUSION

Intestinal AVMs are rare but should be considered as differential diagnoses in patients with recurrent gastrointestinal bleeding. Endoscopic hemostatic, surgical resection, and angiographic intervention can be considered treatments for AVM.

REFERENCES

- Khan MI, Baqai MT, Baqai MF, Mufti N. Exsanguinating upper GI bleeds due to unusual arteriovenous malformation (AVM) of stomach and spleen: A case report. *World Journal of Emergency Surgery*. 2009;4(1).
- Su C, Yang T, Tsai M. A rare cause of gastrointestinal bleeding: Duodenal arteriovenous malformation. *Advances in Digestive Medicine*. 2021;8(1):55–8.
- Kurata Y, Hayano K, Matsusaka K, Mamiya H, Uesato M, Murakami K, et al. A case report of duodenal arteriovenous malformation: usefulness of intraoperative indocyanine green angiography for precise identification of the lesion. *Surg Case Rep*. 2022;8(1):4–9.
- Shrestha S, Pradhan S, KC A, Shrestha S, Kansakar P. Arteriovenous malformation of the jejunum, causing massive gastrointestinal bleeding, treated with intraoperative enteroscopy guidance: A Case Report. *Cureus*. 2023 Jun 4;
- Dunphy L, Ford J. Arteriovenous malformation of the small intestine presenting with a transfusion-dependent anaemia in pregnancy. *BMJ Case Rep*. 2023 Mar 8;16(3).
- Arnautovic JZ, Mazhar A, Tereziu S, Gupta K. A rare association of congenital asplenia with jejunal arteriovenous malformation. Vol. 18, *American Journal of Case Reports*. International Scientific Information, Inc.; 2017. p. 1118–22.
- Havlichek DH 3rd, Kamboj AK, Leggett CL. A Practical Guide to the Evaluation of Small Bowel Bleeding. *Mayo Clin Proc*. 2022;97(1):146-153.
- Trieu DT, Ho AH, Nguyen TTT, Van Le Q. Colorectal arteriovenous malformations causing prolonged bleeding were managed successfully by laparoscopic low anterior resection with sphincter preservation: a case report. *The Korean Journal of Gastroenterology*. 2023 May 25;81(5):216–20.
- Hyo T, Matsuda K, Tamura K, Iwamoto H, Mitani Y, Mizumoto Y, et al. Small intestinal arteriovenous malformation treated by laparoscopic surgery using intravenous injection of ICG: Case report with literature review. *Int J Surg Case Rep*. 2020 Jan 1;74:201–4.
- Ono H, Kusano M, Kawamata F, Danjo Y, Kawakami M, Nagashima K, et al. Intraoperative localization of arteriovenous malformation of a jejunum with combined use of angiographic methods and indocyanine green injection: Report of a new technique. *Int J Surg Case Rep*. 2016;29:137–40.
- Wu LM, Xu JR, Yin Y, Qu XH. Usefulness of CT angiography in diagnosing acute gastrointestinal bleeding: A meta-analysis. *World J Gastroenterol*. 2010;16(31):3957–63.
- Fujii T, Morita H, Sutoh T, Takada T, Tsutsumi S, Kuwano H. Arteriovenous malformation detected by small bowel endoscopy. *Case Rep Gastroenterol*. 2014 Apr 12;8(3):324–8.
- Otani K, Watanabe T, Shimada S, Hosomi S, Nagami Y, Tanaka F, et al. Clinical utility of capsule endoscopy and double-balloon enteroscopy in the management of obscure gastrointestinal bleeding. In: *Digestion*. S. Karger AG; 2018. p. 52–8.
- Sakai E, Ohata K, Nakajima A, Matsushashi N. Diagnosis and therapeutic strategies for small bowel vascular lesions. *World J Gastroenterol*. 2019;25(22):2720–33.
- Mathew Shibi et al. Endosonographic diagnosis and coil embolization of a duodenal arteriovenous malformation. *Endoscopy* 2016; 48: E378–E379
- Bezerra KB, Bacelar Júnior EA, Pereira NC de S, da Costa FA. Gastric arteriovenous malformation: Treatment by embolization. *Radiol Bras*. 2012;45(2):126–8.
- Laskaratos FM, Sehgal V, Haidry R. Successful endoscopic management of a large duodenal arteriovenous malformation using an over-the-scope clip. Vol. 9, *Endoscopy International Open*. Georg Thieme Verlag; 2021. p. E909–10.
- Hirakawa M, Ishizuka R, Sato M, Hayasaka N, Ohnuma H, Murase K, et al. Management of multiple arteriovenous malformations of the small bowel. *Case Rep Med*. 2019;2019.
- Tohma T, Okabe Y, Ushio M, Saito M. Arteriovenous malformation of small intestine successfully treated by double-balloon enteroscopy and laparoscope-assisted surgery. *J Surg Case Rep*. 2022 Dec 1;2022(12).
- McCrary JB, Tang SJ, Wu R, Khangura S, Marks C, Dave B. Endoscopic and angiographic diagnosis and management of a gastric arteriovenous malformation. *Video Journal and Encyclopedia of GI Endoscopy*. 2014;2(1):15–9.
- Gillespie CJ, Sutherland AD, Mossop PJ, Woods RJ, Keck JO, Heriot AG. Mesenteric embolization for lower gastrointestinal bleeding. *Dis Colon Rectum*. 2010 Sep;53(9):1258–64.
- Strate LL, Naumann CR. The role of colonoscopy and radiological procedures in the management of acute lower intestinal bleeding. *Clinical Gastroenterology and Hepatology*. 2010 Apr;8(4):333–43.