



# MID ILEUM ARTERIOVENOUS MALFORMATION : AN UNCOMMON CAUSE OF GASTROINTESTINAL BLEEDING

Aqib Amin<sup>1</sup>, Mudassir Khurshid<sup>2</sup>, Bilal Ahmad<sup>3</sup>, Natasha Thakur<sup>4</sup>, Rayees Fayaz<sup>5</sup>

## ABSTRACT

Bleeding from gastrointestinal tract that originates in the small gut is highly difficult to diagnose such patients usually come with history of obscure gastrointestinal bleed. 65 year old male patient present with a 2 year history of melena and symptomatic transfusion dependent anemia upper and lower GI endoscopy were normal. CT enterography established a great role in source of bleeding as a multiple tortuous arterial phase enhancing vascular channels seen in the gut wall in mid ileum, fed by enlarged branch of SMV. Histological evaluation of the resected gut revealed an arteriovenous malformation. Artificial source light helped in localization of this mass.

**KEYWORDS:** Arteriovenous malformation, ileum, CT enterography.

## INTRODUCTION

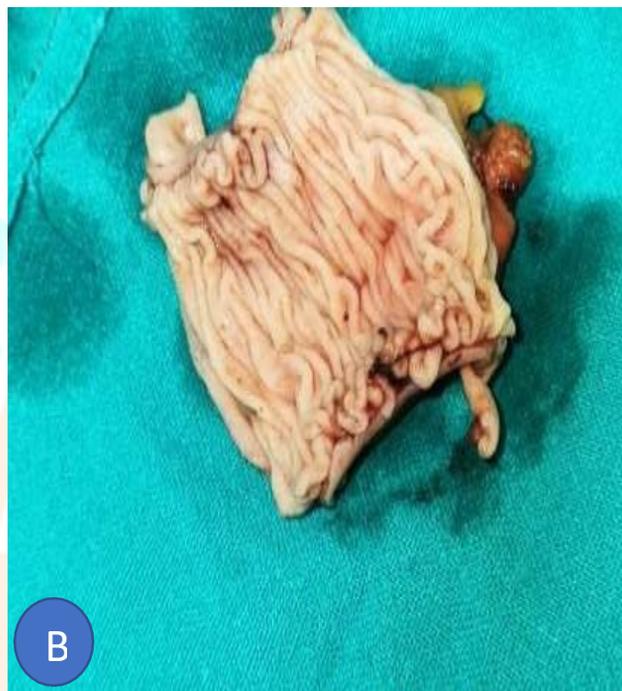
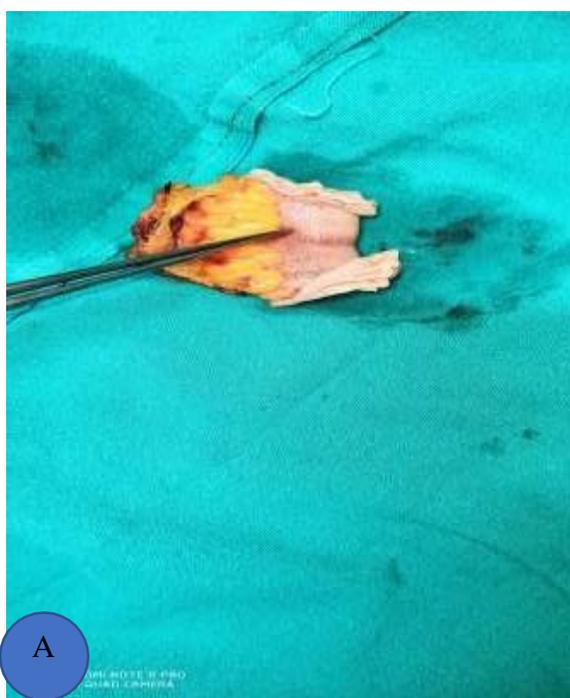
A 65 year old male patient was admitted with, melena that was found to be due to arteriovenous malformation in the mid ileum. The use of CT enterography established the diagnosis before surgical intervention was done and achieving a permanent cure. The importance of this rare case of lower GI bleeding is discussed.

## CASE REPORT

- 65 year old male patient nondiabetic, normotensive, euthyroid was apparently alright 2 years back when he complained of blackish stools and bleeding per rectum. While passing stools, it was mixed with blood most of the times. He also complained of easy fatigability and mild dyspnoea on exertion that had persisted for more than 2 months. His family members noticed that he was becoming progressively pale day by day. He was not taking any medication.
- On examination he was found to be very pale with a pulse of 110 beats/min, blood pressure 110/70. He was afebrile, systemic findings were normal except systolic murmurs. Per rectal and proctoscopic examinations yielded unremarkable findings except for melena.
- Laboratory investigations revealed haemoglobin level of 6 gm/dl, WBC count 8.7 x1000, MCV 77, MCH 20pg, serum iron 21.58, serum ferritin 11, tbc 422, transferrin saturation 51%, ESR 5mm/hr, PLT 166, PT, APTT and INR normal. Stool was positive for occult blood, and CXR and ECG were normal.
- Upper gastrointestinal endoscopy done that showed small gastric polyp. Following this colonoscopy was done upto caecum that didn't show any significant lesion. An emergency CT enterography was done in which axial contiguous were taken through abdomen and pelvis with neutral oral contrast and iv contrast injection in arterial, portal venous and delayed phase images were obtained. Multiple tortuous arterial phase enhancing vascular channels seen in the gut wall in mid ileum, fed by enlarged branch of SMV. The lesion becomes less conspicuous during the portal and delayed phase. These were findings confirming the presence of arteriovenous malformations.

- Patient underwent laparotomy during which a small translucent AVM of ileum (nearly in mid ileum

) was identified .It was very difficult to localise AVM with precise margins during surgery, so we tried to identify avm by illuminating the ileum by narrow beam of torch light extraluminally. Feeding vessels were found that were tortuous at the site of AVM , serosa was slightly reddish (angry looking ) . The AVM along with 7 cm of ileum was resected and end to end anastomosis was done in two layers by vicryl 2 -0 suture .Patient was put on TPN for first 3 days in post surgery period and semisolid food was started on 4<sup>th</sup> post operative day. Histopathological examination of the resected specimen confirmed presence of AVM in ileum. Patient was discharged on 6th day with uneventful recovery. patients haemoglobin continued to drop despite continuous transfusions before surgical intervention , but after surgery his haemoglobin remained 10gm /dl and was still at this level at the time of discharge .



*Figure: Representative images of arteriovenous malformation.*

*A & B: Surgical resection of ileum having AVM*

*C: Pathological Examination revealed a vascular malformation in the submucosa*

*D: CT Enterography showing Mid Ileum AVM*

## DISCUSSION

- Vascular ectasia, which is also termed “angiodysplasia,” “AVM,” and “telangiectasia,” is a relatively uncommon cause of gastrointestinal bleeding [1]. Ectasia may be found anywhere in the gastrointestinal tract, from the nasopharynx to the rectum [2-5]. It may never bleed and be discovered only at autopsy [6]. The most common site of AVM in the gastrointestinal tract is the caecum and the right colon and represents about 77.5% of cases. Ileal AVMs constitute only 5.5 % of total cases [7].

- It might be challenging to make a diagnosis of gastrointestinal bleeding that starts in the small intestine [8 – 12].

- The term "obscure gastrointestinal bleeding" (OGIB) refers to gastrointestinal (GI) bleeding from an undetermined source that persists despite a thorough upper and lower GI evaluation. Most bleeding causes are apparently found in the small intestine, accounting for about 5% of all occurrences of GI bleeding[14]. Missed lesions discovered during an esophagogastroduodenoscopy or colonoscopy should be taken into consideration.

- Small intestinal bleeding can be overt, show up as clinically obvious melena, hematochezia, or occult bleeding,

and be linked to iron-deficiency anemia with or without a positive faecal occult blood test[16].

- The GI tract has been linked to several different vascular disorders. The most prevalent cause of small bowel bleeding, angiodysplasia (AD), is characterized by a focused buildup of aberrant, dilated, and convoluted blood vessels visible within the mucosal and submucosal layers of the gut.[17 ,18]. Patients with OGIB are typically found to have AD, and the degree of bleeding can vary

greatly from chronic, well-tolerated situations to acute life-threatening conditions[19].

- Based on the Yano-Yamamoto classification, small bowel vascular lesions can be endoscopically divided into four categories[20]. Type 1a: Punctuate ( 1 mm) or Type 1b: Patchy (a few mm) AEs are typically identified during endoscopy as tiny erythemas. They are weak and prone to bleeding because they are histopathologically made up of narrow, dilated, and convoluted veins that lack a smooth muscle layer. In Type 2a DLs, excessively big arteries that are histologically normal yet protrude via a tiny mucosal defect: With pulsatile bleeding or Type 2b: Pulsatile red protrusions without surrounding venous dilatation, punctuate lesions[21] . AVMs are characterized by the direct connections of arteries and veins without a capillary bed and are histopathologically recognized as aberrant vessels with thicker, hypertrophic walls that vary in thickness greatly[22].

- Type 3: Pulsatile red protrusions with surrounding venous dilatation, describes some intestinal AVMs. Congenital intestinal AVMs, on the other hand, can occasionally manifest as a mass or polypoid lesion[23,24] and are therefore classed as Type 4: Lesions not falling under any of the aforementioned classifications.

- According to Boley et al. [25], persistent hypoxia and raised intestinal wall pressures might cause submucosal veins to partially occlude, which can cause capillary congestion, pre-capillary sphincter failure, and eventually the development of permanent AE. The fact that AE is usually found in the right colon of older individuals, where bowel tension is quite considerable, supports this theory[26–28].

- Angiogenic factors are crucial in the development of AE, according to Junquera et al. They discovered that patients with colonic AEs had considerably higher expression levels of vascular endothelial growth factor (VEGF), a key mediator in the early stages of angiogenesis. According to reports, the balance between pro- and anti- angiogenic factors is apparently disrupted by persistent hypoxia-induced mucosal ischemia, which can be caused in part by cardiac or renal disorders and lead to pathological neovascularization[30–31].

- The diagnosis and management of small bowel ADs involve the use of deep enteroscopy (DE) and video capsule endoscopy (VCE). These modalities have different traits despite having the same indications for usage. In about 90% of individuals, VCE makes the entire small bowel visible[32], and the usage of real-time watching or a larger battery can raise this success percentage even more[33]. When used to detect continuous overt bleeding, VCE had the highest diagnostic yield[34]. , It shows how useful emergent VCE is. Be aware that emergent VCE can be used to choose the management strategy after determining the cause of bleeding. According to reports, VCE is more effective than other diagnostic modalities for detecting ADs, including computed tomography (CT) enterography, mesenteric angiography, and DE[35]. In order to determine whether small bowel bleeding is present, GI organizations presently advise using VCE as a first-line test[36]. But there are still some issues with VCE's restrictions. The biggest drawback is its inability to gather biopsy samples or perform endoscopic procedures.

- For patients with active overt GI bleeding, radiographic investigations such as multiphase CT angiography, radionuclide scanning, and mesenteric angiography are helpful in identifying the bleeding source. The bleeding rate threshold and the intermittent pattern of small bowel AD bleeding should be taken into account when interpreting the results of radiographic investigations. When the bleeding rate exceeds 0.3 mL/min in patients with overt GI bleeding, multi-phase CT angiography can precisely localize the bleeding region as an extravasation[35].

- When the bleeding rate exceeds 0.1 mL/min, localizing the bleeding source can be done by radionuclide scanning utilizing technetium-99m-labeled red blood cells. According to some sources, up to 66% of positive test results are accurate[37].

- Despite its noninvasiveness and sensitivity in detecting bleeding, radionuclide scanning has problems pinpointing the exact location of the bleeding site. Furthermore, this method is limited to diagnostic application; therefore, a subsequent endoscopic or angiographic examination is necessary. Mesenteric angiography has a rather low sensitivity[38]. Because in order to allow for diagnosis and therapy, an active bleeding rate of more than 0.5 mL/min must be present at the time of the examination. However, this method enables precise localization and subsequent selective embolization during the same examination. The rate of bleeding plays a key role in the successful localization of bleeding, which may be especially helpful for patients with hemodynamic instability who need big blood transfusions.[39] .

- Similar to DLs, intestinal AVMs can potentially result in fatal bleeding[40]. Despite their relatively low occurrence, small bowel AVMs can be used to pinpoint the bleeding source in patients with overt OGIB who also have severe, transfusion-dependent anemia. The majority of AVM causes are congenital, hence younger patients need to be treated with extra caution. VCE is a practical diagnostic tool for locating small bowel AVMs. CT enterography is also advised for identifying small bowel AVMs due to the high detection rate.[41] .

- Small AVMs that endoscopically appear as flat or slightly elevated hemorrhagic patches might be treated with endoclips to achieve mechanical hemostasis during successive DE examinations, just as DLs.[20]. However, due to their often enormous size and propensity to rebleed, the majority of small intestinal AVMs need to be surgically removed. It can be challenging to locate the lesion during surgery for patients with small intestinal AVMs.[42] . For primary hemostasis and preoperative localization, mesentery angiography and subsequent microcoil embolization are allegedly effective[42]. Furthermore, it has been claimed that intraoperative indocyanine green

injections[40] and endoscopic tattooing[23] or marking clips are effective for locating the target lesion.

## REFERENCES

1. Levine DS. Intestinal vascular ectasis: improving diagnostic capability poses therapeutic dilemma. *Am J Med* 1984;76:1151-5.
2. Al-Humadi AH, Domaleski AF, Alford JE, et al. Arteriovenous malformation of the upper rectum in an unusual site: report of a case and review of the literature. *Dis Colon Rectum* 1979;22:189-94.
3. Farup PG, Rosseland AR, Stray N, et al. Localized telangiopathy of the stomach and duodenum diagnosed and treated endoscopically: case reports and review. *Endoscopy* 1981;13:1-6.
4. Gagie N, Thurlow W. Arteriovenous malformation of the jejunum. *Can Med Assoc J* 1977;117:729-30.
5. Ottinger LW, Vickery AL Jr. A thirty year history of recurrent gastrointestinal bleeding. *N Engl J Med* 1981;305:211-6.
6. Boley JJ, Sammartano R, Brandt LJ, et al. Vascular ectasias of the colon. *Surg Gynecol Obstet* 1979;149:353-9.
7. Meyer CT, Troncale FJ, Galloway S, Sheahan DG. Arteriovenous malformations of the bowel: an analysis of 22 cases and a review of the literature. *Medicine* 1981;60(1):36-48.
8. Endo H, Matsuhashi N, Inamori M, Ohya T, Yanagawa T, Asayama M, Hisatomi K, Teratani T, Fujita K, Yoneda M, Nakajima A: Tumorous arteriovenous malformation in the jejunum missed by capsule endoscopy. *Gastrointest Endosc* 2008;68:773-774; discussion 774.
9. Rizvi AZ, Kaufman JA, Smith P, Silen ML: Solitary arteriovenous malformation of the small intestine. *J Am Coll Surg* 2005;200:808-809.
10. Stauffer JA, Shaddix KK, Achem SR, Stark M, Adelson A, Metzger PP, Landmann RG: Intra-operative use of super-selective or highly selective angiography with methylene blue injection to localize arterial-venous malformation. *Colorectal Dis* 2011;13:e65-e66.
11. Takeshita N, Otsuka Y, Nara S, Noie T, Ito K, Harihara Y, Furushima K, Konishi T: Utility of preoperative small-bowel endoscopy for haemorrhagic lesions in the small intestine. *Surg Today* 2012;42:536-541.
12. Nakabayashi T, Kudo M, Hirasawa T, Kuwano H: Arteriovenous malformation of the jejunum detected by arterial-phase enhanced helical CT, a case report. *Hepatogastroenterology* 2004;51:1066-1068.
13. ASGE Standards of Practice Committee; Gurudu SR, Bruining DH, Acosta RD, Eloubeidi MA, Faulx AL, Khashab MA, Kothari S, Lightdale JR, Muthusamy VR, Yang J, DeWitt JM. The role of endoscopy in the management of suspected small-bowel bleeding. *Gastrointest Endosc* 2017; 85: 22-31 [PMID: 27374798 DOI: 10.1016/j.gie.2016.06.013]
14. Gerson LB, Fidler JL, Cave DR, Leighton JA. ACG Clinical Guideline: Diagnosis and

## Management of Small Bowel

- Bleeding. *Am J Gastroenterol* 2015; 110: 1265-87; quiz 1288 [PMID: 26303132 DOI: 10.1038/ajg.2015.246]
15. Lewis BS. Small intestinal bleeding. *Gastroenterol Clin North Am* 1994; 23: 67-91 [PMID: 8132301 DOI: 10.1016/S0889-8553(05)70108-4] Eiji
16. Sakai, Ken Ohata, Nobuyuki Matsuhashi, *World J Gastroenterol* 2019 June 14; 25(22): 2720-2733.
17. Li F, Leighton JA, Sharma VK. Capsule endoscopy in the evaluation of obscure gastrointestinal bleeding: A comprehensive review. *Gastroenterol Hepatol (N Y)* 2007; 3: 777-785 [PMID: 21960786]
18. Sakai E, Endo H, Taniguchi L, Hata Y, Ezuka A, Nagase H, Yamada E, Ohkubo H, Higurashi T, Sekino Y, Koide T, Iida H, Hosono K, Nonaka T, Takahashi H, Inamori M, Maeda S, Nakajima A. Factors predicting the presence of small bowel lesions in patients with obscure gastrointestinal bleeding. *Dig Endosc* 2013; 25: 412-420 [PMID: 23368528 DOI: 10.1111/den.12002]
19. Almeida N, Figueiredo P, Lopes S, Freire P, Lérias C, Gouveia H, Leitão MC. Urgent capsule endoscopy is useful in severe obscure-overt gastrointestinal bleeding. *Dig Endosc* 2009; 21: 87-92 [PMID: 19691780 DOI: 10.1111/j.1443-1661.2009.00838]
20. Yano T, Yamamoto H, Sunada K, Miyata T, Iwamoto M, Hayashi Y, Arashiro M, Sugano K. Endoscopic classification of vascular lesions of the small intestine (with videos). *Gastrointest Endosc* 2008; 67: 169-172 [PMID: 18155439 DOI: 10.1016/j.gie.2007.08.005]
21. Morowitz MJ, Markowitz R, Kamath BM, von Allmen D. Dieulafoy's lesion and segmental dilatation of the small bowel: An uncommon cause of gastrointestinal bleeding. *J Pediatr Surg* 2004; 39: 1726-1728 [PMID: 15547843 DOI: 10.1016/j.jpedsurg.2004.07.027]
22. Eastman J, Nazek M, Mangels D. Localized arteriovenous malformation of the jejunum. *Arch Pathol Lab Med* 1994; 118: 181-183 [PMID: 8311661 DOI: 10.1002/sim.4780130310]
23. Chung CS, Chen KC, Chou YH, Chen KH. Emergent single-balloon enteroscopy for overt bleeding of small intestinal vascular malformation. *World J Gastroenterol* 2018; 24: 157-160 [PMID: 29358892 DOI: 10.3748/wjg.v24.i1.157]
24. Molina AL, Jester T, Nogueira J, CaJacob N. Small intestine polypoid arteriovenous malformation: A stepwise approach to diagnosis in a paediatric case. *BMJ Case Rep* 2018; 2018: pii: bcr-2018-224536 [PMID: 30042105 DOI: 10.1136/bcr-2018-224536]
25. Boley SJ, Sammartano R, Adams A, DiBiase A, Kleinhaus S, Sprayregen S. On the nature and etiology of vascular ectasias of the colon. Degenerative lesions of aging. *Gastroenterology* 1977; 72: 650-660 [PMID: 300063]
26. Danesh BJ, Spiliadis C, Williams CB, Zambartas CM. Angiodysplasia--an uncommon cause of colonic bleeding: Colonoscopic evaluation of 1,050 patients with rectal bleeding and anaemia. *Int J Colorectal Dis* 1987; 2: 218-222 [PMID: 3500991 DOI: 10.1007/BF01649509]
27. Foutch PG, Rex DK, Lieberman DA. Prevalence and natural history of colonic angiodysplasia among healthy asymptomatic people. *Am J Gastroenterol* 1995; 90: 564-567 [PMID: 7717311]
28. Höchter W, Weingart J, Kühner W, Frimberger E, Ottenjann R. Angiodysplasia in the colon and rectum. Endoscopic morphology, localisation and frequency. *Endoscopy* 1985; 17: 182-185 [PMID: 3876926 DOI: 10.1055/s-2007-1018495]
29. Junquera F, Saperas E, de Torres I, Vidal MT, Malagelada JR. Increased expression of

angiogenic factors in human colonic angiodysplasia. *Am J Gastroenterol* 1999; 94: 1070-1076 [PMID: 10201485 DOI: 10.1111/j.1572-0241.1999.01017.x]

30. Galanopoulos G. Angiodysplastic lesions as a cause of colonic bleeding in patients with chronic renal disease: Is there an association? *Saudi J Kidney Dis Transpl* 2012; 23: 925-928 [PMID: 22982901 DOI: 10.4103/1319-2442.100858]

31. Pate GE, Mulligan A. An epidemiological study of Heyde's syndrome: An association between aortic stenosis and gastrointestinal bleeding. *J Heart Valve Dis* 2004; 13: 713-716 [PMID: 15473467]

32. Pennazio M, Santucci R, Rondonotti E, Abbiati C, Beccari G, Rossini FP, De Franchis R. Outcome of patients with obscure gastrointestinal bleeding after capsule endoscopy: Report of 100 consecutive cases. *Gastroenterology* 2004; 126: 643-653 [PMID: 14988816 DOI: 10.1053/j.gastro.2003.11.057]

33. Hadithi M, Heine GD, Jacobs MA, van Bodegraven AA, Mulder CJ. A prospective study comparing video capsule endoscopy with double-balloon enteroscopy in patients with obscure gastrointestinal bleeding. *Am J Gastroenterol* 2006; 101: 52-57 [PMID: 16405533 DOI: 10.1111/j.1572-0241.2005.00346.x]

34. Pennazio M, Spada C, Eliakim R, Keuchel M, May A, Mulder CJ, Rondonotti E, Adler SN, Albert J, Baltes P, Barbaro F, Cellier C, Charton JP, Delvaux M, Despott EJ, Domagk D, Klein A, McAlindon M, Rosa B, Rowe G, Sanders DS, Saurin JC, Sidhu R, Dumonceau JM, Hassan C, Gralnek IM. Small-bowel capsule endoscopy and device-assisted enteroscopy for diagnosis and treatment of small-bowel disorders: European Society of Gastrointestinal Endoscopy (ESGE) Clinical Guideline. *Endoscopy* 2015; 47: 352-376 [PMID: 25826168 DOI: 10.1055/s-0034-1391855]

35. Ren JZ, Zhang MF, Rong AM, Fang XJ, Zhang K, Huang GH, Chen PF, Wang ZY, Duan XH, Han XW, Liu YJ. Lower gastrointestinal bleeding: Role of 64-row computed tomographic angiography in diagnosis and therapeutic planning. *World J Gastroenterol* 2015; 21: 4030-4037 [PMID: 25852291 DOI: 10.3748/wjg.v21.i13.4030]

36. Huprich JE, Barlow JM, Hansel SL, Alexander JA, Fidler JL. Multiphase CT enterography evaluation of small-bowel vascular lesions. *AJR Am J Roentgenol* 2013; 201: 65-72 [PMID: 23789659 DOI: 10.2214/AJR.12.10414]

37. Strate LL, Naumann CR. The role of colonoscopy and radiological procedures in the management of acute lower intestinal bleeding. *Clin Gastroenterol Hepatol* 2010; 8: 333-43; quiz e44 [PMID: 20036757 DOI: 10.1016/j.cgh.2009.12.017]

38. Leung WK, Ho SS, Suen BY, Lai LH, Yu S, Ng EK, Ng SS, Chiu PW, Sung JJ, Chan FK, Lau JY. Capsule endoscopy or angiography in patients with acute overt obscure gastrointestinal bleeding: A prospective randomized study with long-term follow-up. *Am J Gastroenterol* 2012; 107: 1370-1376 [PMID: 22825363 DOI: 10.1038/ajg.2012.212]

39. Abbas SM, Bissett IP, Holden A, Woodfield JC, Parry BR, Duncan D. Clinical variables associated with positive angiographic localization of lower gastrointestinal bleeding. *ANZ J Surg* 2005; 75: 953-957 [PMID: 16336385 DOI: 10.1111/j.1445-2197.2005.03582.x]

40. Ono H, Kusano M, Kawamata F, Danjo Y, Kawakami M, Nagashima K, Nishihara H. 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: 137-140 [PMID: 27846454 DOI: 10.1016/j.ijscr.2016.10.030].

41. Huprich JE, Fletcher JG, Fidler JL, Alexander JA, Guimarães LS, Siddiki HA, McCollough CH. Prospective blinded comparison of wireless capsule endoscopy and multiphase CT enterography in obscure gastrointestinal bleeding. *Radiology* 2011; 260: 744-751 [PMID: 21642417 DOI: 10.1148/radiol.11110143]

42. So M, Itatani Y, Obama K, Tsunoda S, Hisamori S, Hashimoto K, Sakai Y. Laparoscopic resection of idiopathic jejunal arteriovenous malformation after metallic coil embolization. *Surg Case Rep* 2018; 4: 78 [PMID: 30022275 DOI: 10.1186/s40792-018-0486-4].