Philippine Journal of Gastroenterology

A rare case of lower gastrointestinal bleeding from an appendiceal arteriovenous malformation in a Filipino

[1]Tan, KC

[2]Lim, CC

[2]Ong, EG

[2]Ong-Go, AK

[1]Department of Internal Medicine

[2]Department of Internal Medicine - Section of Gastroenterology

Metropolitan Medical Center
1357 Masangkay St., Sta. Cruz
Manila 1008, Philippines

Correspondence:
Karla Chrizelle Tan, MD
tan.karlachrizelle@gmail.com

Abstract

Background: Acute bleeding from the appendix is uncommon with only 34 reported cases in the literature. To our knowledge, this is the first reported case in the Philippines of gastrointestinal bleeding secondary to an arteriovenous malformation of the appendix. This case highlights the importance of including the appendix as a possible source of lower gastrointestinal bleeding.

Case Summary: A 32-year-old male presented with acute painless hematochezia. On urgent colonoscopy, fresh and dark blood with clots filled the entire colon, while the terminal ileum was free from blood which was highly suggestive of a colonic source of bleeding. After a thorough irrigation of the ileocecal region, significant amount of blood was seen oozing intermittently from the appendiceal orifice. An emergency laparoscopic appendectomy was performed. On gross examination, the appendix was enlarged and on cut section, there was a prominent vessel. Histopathologic examination showed acute appendicitis with serosal congestion and there were thickened and enlarged vessels. Immunohistochemistry was positive for CD31 and CD34, consistent with arteriovenous malformation.

Conclusion: In cases of suspected colonic bleeding with no obvious cause, appendiceal hemorrhage should be one of the considerations so as not to miss this rare but treatable cause of lower gastrointestinal bleeding.

Keywords: *Appendix bleeding, Appendix hemorrhage, Arteriovenous malformation*

Introduction

Lower gastrointestinal bleeding (LGIB) is a serious and common condition that can be life-threatening, often caused by bleeding from a source distal to the ligament of Treitz. Lower gastrointestinal bleeding accounts for 20% of gastrointestinal bleeding.[1] Once stabilized, patients with lower gastrointestinal bleeding should undergo urgent colonoscopy.[1] Identifying the source of bleeding is a clinical priority, and can be challenging when compared to upper gastrointestinal bleeding.[2] In 10% of cases, the bleeding source cannot be identified even with extensive evaluation.[3] Appendiceal bleeding constitutes approximately 0.014% of gastrointestinal bleeding cases.[4] In this paper, we present a rare case of lower gastrointestinal bleeding from an appendiceal arteriovenous (AV) malformation.

Case Presentation

A previously well 32-year-old Filipino male, presented with painless hematochezia eight hours prior to consult. He passed out fresh and dark blood thrice, around 100-200 ml per bout. He had no known illnesses and denied taking any medications. He did not smoke cigarettes or consume alcohol. The patient was conscious and coherent, with stable vital signs; abdominal examination and laboratory results were normal.

On urgent colonoscopy, blood was encountered in the entire colon. No source of bleeding was found in the colon despite vigorous flushing and cleaning. The terminal ileum was intubated to around 30 cm from the ileocecal valve. The ileal mucosa was unremarkable and contained some yellow stools with no traces of blood. As the scope was being withdrawn, there was pooling of fresh blood in the cecal pole. After cleaning the area, blood was seen oozing intermittently from the appendiceal orifice (Figures 1 and 2).

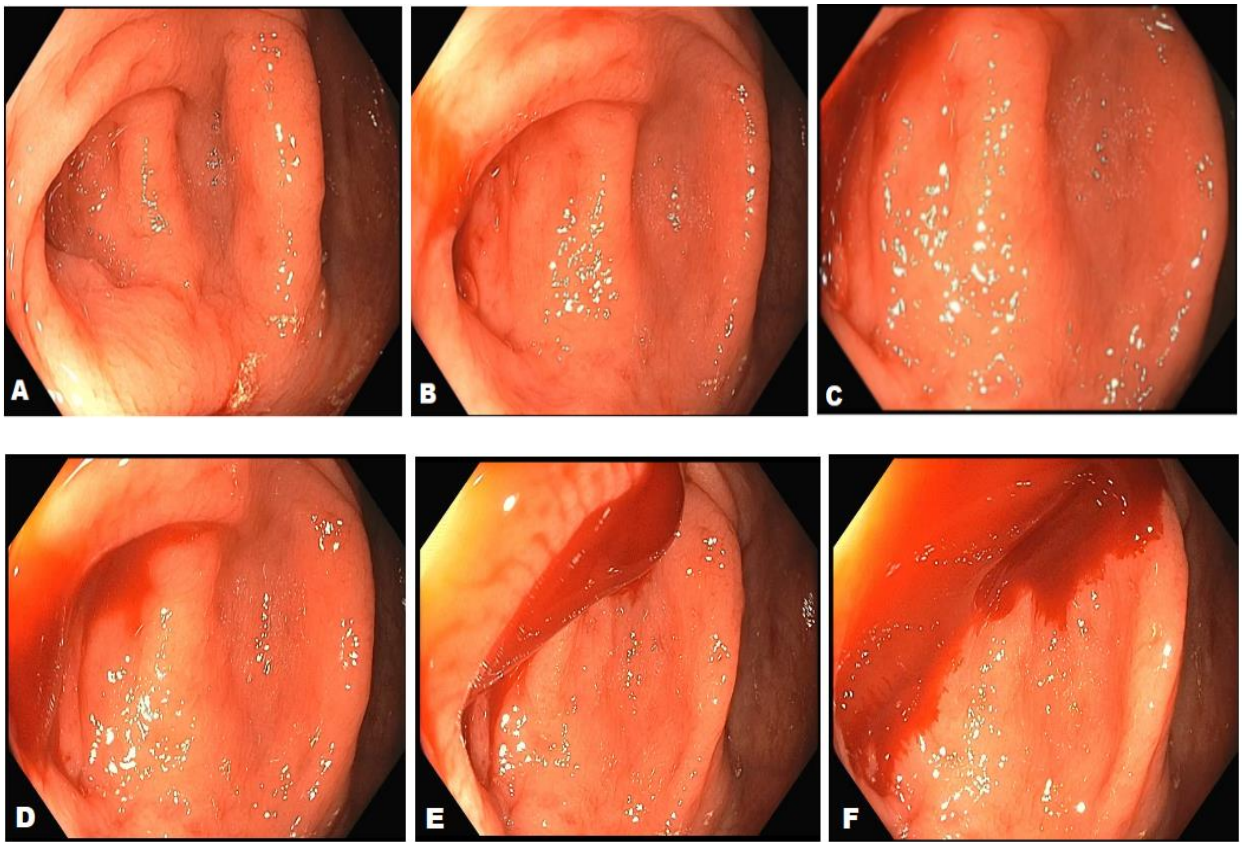

Figure 1. A to F: Sequential images of blood oozing from the appendiceal orifice.

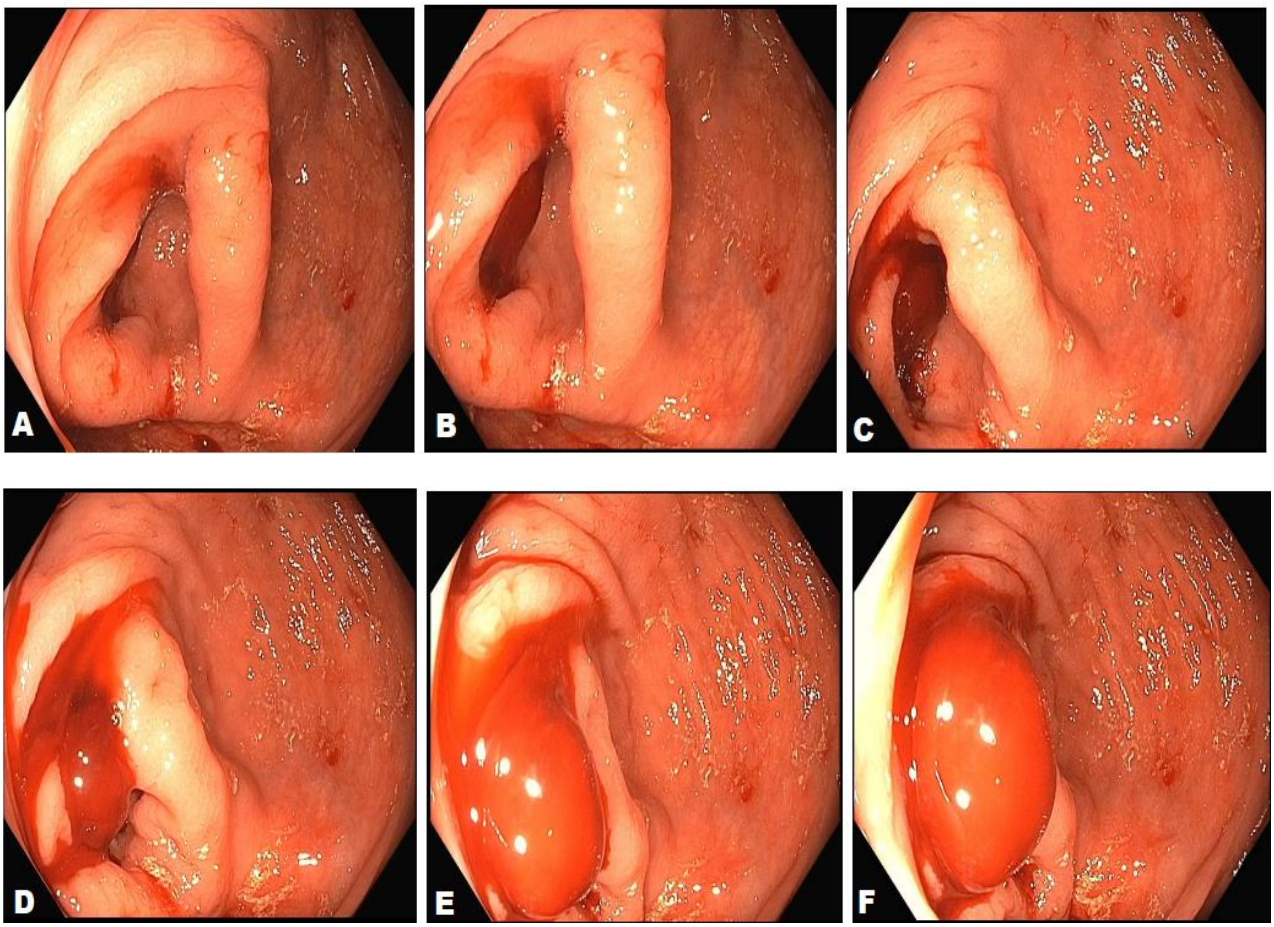

Figure 2. A to F: Sequential images showing intermittent extrusion of fresh blood and clots from the appendiceal opening after repeated cleaning by water flushing and suctioning.

The patient then underwent emergency laparoscopic appendectomy. The appendix was retrocecal and had minimal adhesions to the right abdominal wall (Figure 3). There were no indications of rupture, diverticula, nor intestinal bands.

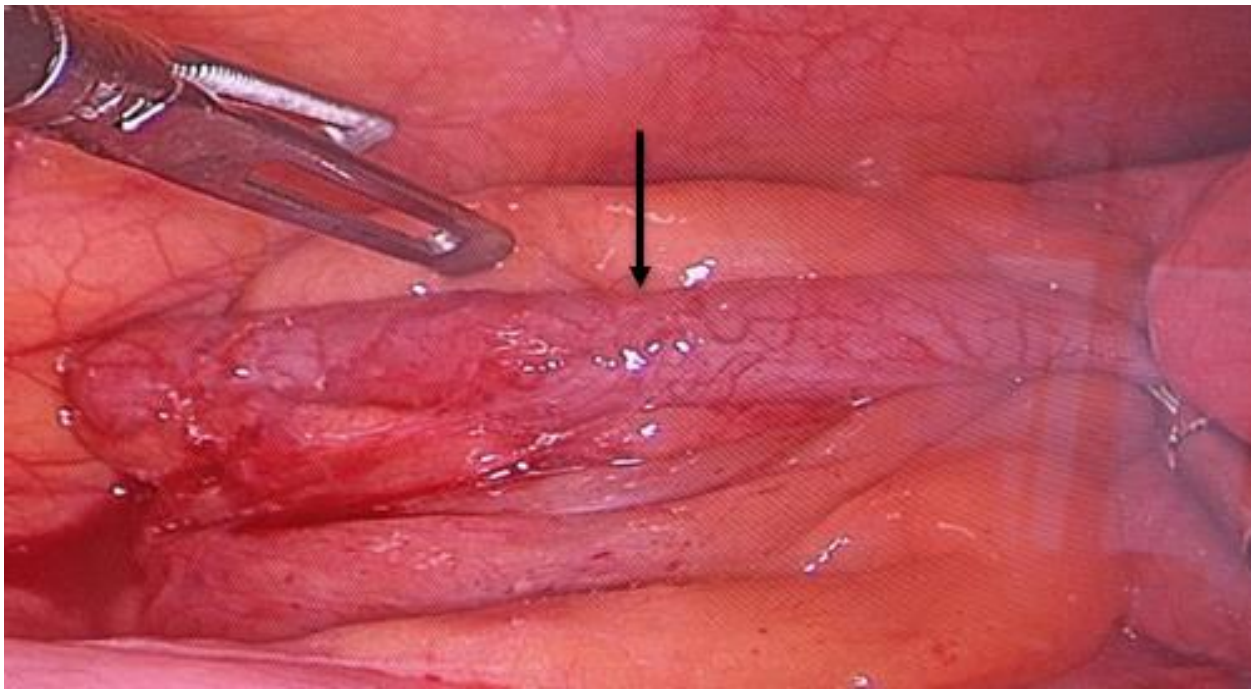

Figure 3. Arrow pointing at a laparoscopic image showing an enlarged, retrocecal appendix.

On gross examination, the resected appendix measured 4.7 cm x 1.5 cm x 0.5 cm in size with serosal congestion (Figure 4A). Cut section showed a prominent vessel (Figure 4B).

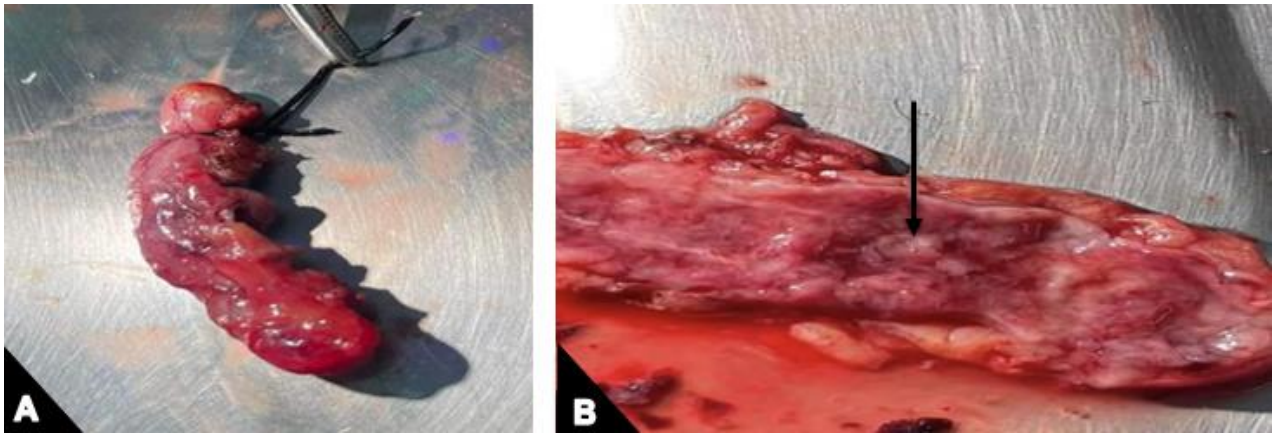

Figure 4. A: Resected appendix. B: Cut specimen showed a tan surface with a prominent vessel (arrow) measuring around 2mm in diameter.

Histopathologic examination showed acute appendicitis with serosal congestion (Figure 5A). There were thickened and enlarged vessels on the mucosa (Figure 5B).

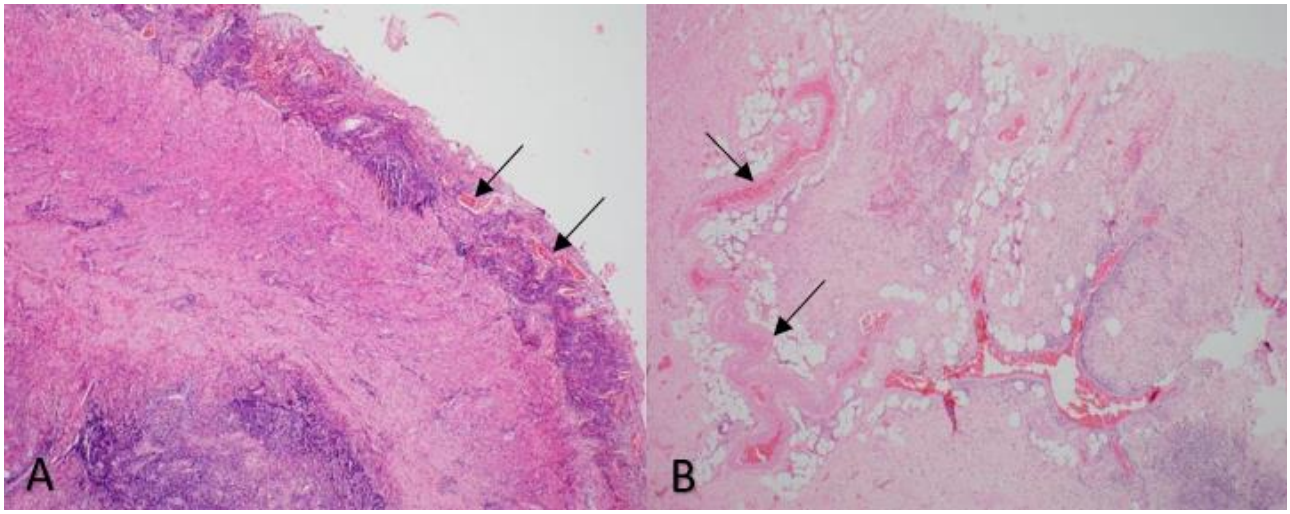

Figure 5. A: Arrows show Hematoxylin and Eosin (H&E) stain demonstrating appendiceal serosa with thin-walled blood vessels containing red blood cells indicating congestion. B: Arrows show enlarged and thickened blood vessels on the mucosa ascribing to hemangioma or AV malformation.

The immunohistochemistry showed positive results for CD31 and CD34 which were expressions on the membrane of endothelial cells of malformed vessels. These findings were consistent with AV malformations (Figures 6A and 6B).

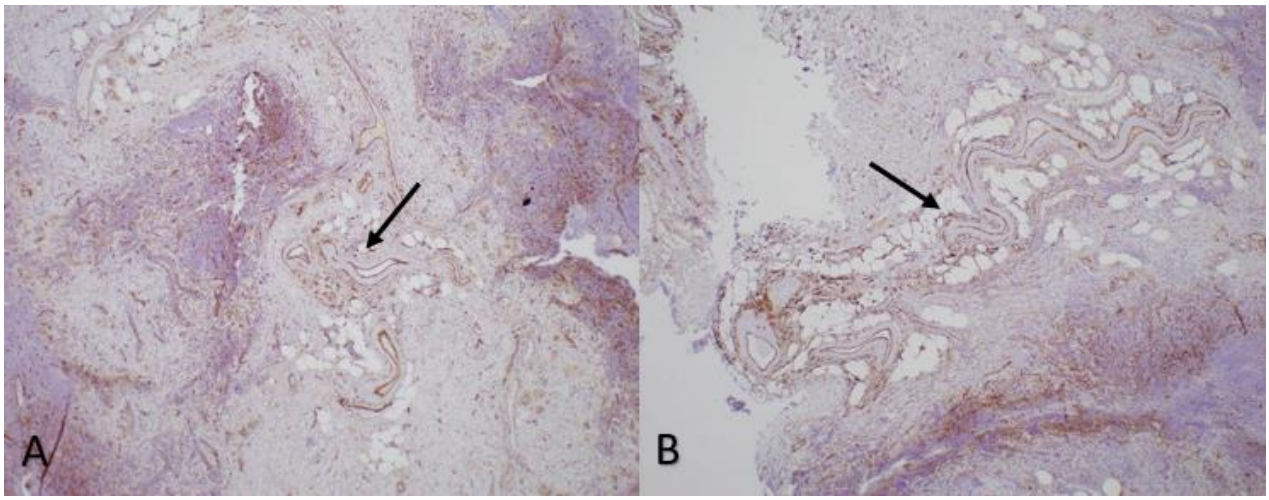

Figure 6. A and B: Arrows indicate Immunohistochemical staining for endothelial cells highlighting blood vessels.

The postoperative course was uneventful and the patient did not require blood transfusion. On follow-up, the patient was well and there was no recurrence of hematochezia.

Discussion

Lower gastrointestinal bleeding (LGIB) is a common cause of hospitalization. The majority of LGIB originate from the colon, with a small percentage arising from the small bowel located distal to the ligament of Treitz. Patients with LGIB usually require blood transfusions and interventions such as gastrointestinal endoscopy and surgical treatment.[5] Common causes of colonic bleeding include diverticular bleeding, angioectasias, post-polypectomy bleeding, and ischemic colitis.[1, 6] Other less common causes include rectal ulcers, infectious colitis, inflammatory bowel disease, colorectal polyps and neoplasms, radiation proctitis, and hemorrhoids.[1, 5, 6] However, bleeding from the appendix has limited literature.

A search on PubMed/MEDLINE database for literature published between January 1977 to September 2024 regarding "appendix bleeding" or "appendix hemorrhage" identified 34 articles.

Table 1. Summary of the articles published on appendiceal bleeding.

No	Year [Ref]	Age/ Sex	Clinical Presentation	Diagnostic Modality	Diagnosis	Treatment
1	2024 [7]	33/F	Hematochezia	Colonoscopy	Appendiceal AV malformation	Laparoscopic appendectomy
2	2024 [8]	79/F	Hematochezia	CT Angiography	Appendiceal ulcer	Appendectomy
3	2023 [9]	32/M	Hematochezia	Operative	Appendiceal ulcer	Endoscopic appendectomy
4	2023 [10]	57/M	Hematochezia	Angiography	Mesoappendiceal AV malformation	Laparoscopic appendectomy
5	2022 [11]	32/M	Hematochezia	Colonoscopy	Appendiceal Dieulafoy lesion	Laparoscopic appendectomy
6	2021 [12]	90/M	Melena	Colonoscopy	Appendiceal wall bleed	Laparoscopic appendectomy
7	2021 [13]	67/M	Hematochezia	Mesenteric angiogram	Appendiceal artery bleed	Right hemicolectomy
8	2021 [14]	82/M	Hematochezia	Colonoscopy	Acute Eosinophilic Appendicitis	Laparoscopic partial cecectomy
9	2021 [15]	53/M	Hematochezia	Colonoscopy	Appendiceal AV malformation	Appendectomy
10	2020 [16]	88/M	Hematochezia	Operative	Low-Grade Mucinous Neoplasm of Appendix	Appendectomy
11	2019 [17]	24/M	Hematochezia	Colonoscopy	Granulomatous appendicitis	Appendectomy
12	2019 [18]	67/M	Melena	Operative	Iliac-Appendiceal fistula	Appendectomy and partial cecectomy
13	2017 [19]	46/M	Melena	Operative	Appendicitis	Laparoscopic appendectomy
14	2016 [20]	72/M	Hematochezia	Operative, Microscopic	Angiodysplasia	Laparoscopic appendectomy, cecum wedge resection
15	2016 [21]	22/M	Hematochezia	CT angiography	Granulomatous Appendicitis	Appendectomy
16	2015 [22]	68/M	Hematochezia	Colonoscopy	Appendiceal Dieulafoy lesion	Laparoscopic appendectomy
17	2014 [23]	44/M	Hematochezia	Colonoscopy	Appendiceal Diverticulum	Right hemicolectomy

No	Year [Ref]	Age/ Sex	Clinical Presentation	Diagnostic Modality	Diagnosis	Treatment
18	2014 [24]	51/M	Appendicitis	Operative	Appendiceal Dieulafoy lesion	Appendectomy
19	2013 [25]	71/M	Melena	Colonoscopy	Appendiceal ulcer	Barium enema, Appendectomy
20	2013 [26]	41/M	Melena	Colonoscopy	Atypical florid vascular proliferations	Laparoscopic appendectomy
21	2012 [27]	59/F	Hematochezia	Operative	Aorto-enteric fistula	Extended right hemicolectomy, femoro-femoral saphenous vein bypass
22	2011 [28]	70/M	Hematochezia	Colonoscopy	Appendiceal orifice bleed	Endoclipping
23	2011 [29]	33/M	Hematochezia	Colonoscopy	Appendiceal mucosal erosion	Appendectomy
24	2011 [30]	25/M	Hematochezia	Multidetector CT	Focal erosion of appendix mucosa	Appendectomy
25	2010 [31]	34/F	Melena	Colonoscopy	Appendiceal orifice bleed	Colonoscopic clipping
26	2010 [32]	42/M	Hematochezia	Multidetector helical CT, Colonoscopy	Appendiceal mucosal erosion	Laparoscopic appendectomy
27	2007 [33]	56/M	Hematochezia	Colonoscopy	Gastrointestinal Stromal Tumor (GIST)	Right hemicolectomy with appendectomy
28	2004 [34]	16/M	Hematochezia	Colonoscopy	Crohn's Disease	Appendectomy
29	2003 [35]	33/M	Hematochezia	Colonoscopy	Appendiceal stump bleed	Colonoscopic hemoclipping post-appendectomy
30	2001 [36]	76/F	Hematochezia	Mesenteric arteriography	Angiodysplasia	Appendectomy
31	1985 [37]	32/F	Hematochezia	Colonoscopy	Ulcerated appendiceal stump	Right hemicolectomy
32	1980 [38]	48/M	Hematochezia	Mesenteric arteriography	Appendiceal diverticulum	Appendectomy
33	1979 [39]	63/M	Hematochezia	Operative	Appendiceal mucocele	Right colectomy with ileotransverse colostomy and two-layer, end-to-end anastomosis
34	1977 [40]	14/M	Blood clots and pus	Operative	Appendiceal abscess	Abscess drainage and ileocecal resection with anastomosis

Based on the available case reports, appendiceal bleeding was attributed to a variety of etiology including appendicitis, erosion, ulcer and various vascular causes (Table 2).

Table 2. Summary of the causes of appendiceal bleeding.

Diagnosis	Number of cases
Vascular	
• Dieulafoy	3
• Angiodysplasia	2
• AV malformation	3
• Artery bleed	1
• Vascular proliferation	1
Mucosal Break	
• Erosion	3
• Ulcer	4
Inflammatory/Infectious	
• Appendicitis	4
• Abscess	1
Fistula	
• Aorto-Enteric	1
• Iliac-Appendiceal	1
Others	
• Diverticulum	2
• Gastrointestinal Stromal Tumor	1
• Crohn's Disease	1
• Tumor	1
• Mucocele	1
• Wall bleeding	1
• No histopathology	3
TOTAL	34

The mean age of cases of appendiceal bleeding was 50 years (14-90 years). Our patient was 32 years old. The majority of the cases were males (82.3%), and most presented with hematochezia (76%), similar to our case. Half of the cases were diagnosed through colonoscopy (50%). Vascular etiology for bleeding accounted for 29.41% of the cases.

Identifying the source of LGIB could be a challenge. Some cases would require the use of more than one modality to arrive at a diagnosis. This was also the case among published papers of appendiceal bleeding. In up to 10% of cases of LGIB, no source could be identified despite extensive work up. Fortunately for our case, the bleeding source was identified during colonoscopy and no further work-up was needed.

There were only three reported cases of appendiceal AV malformation. One case presented with a hemoglobin level within the normal range, like in our case, while two presented with anemia. One had a colonoscopy showing continuous bleeding at the appendiceal orifice followed by a contrast-enhanced CT finding of a structural disorder in the ileocecal area.[7] Another case was initially noted to have a thickened appendix without signs of inflammation with findings of vascular proliferation and dilated veins on contrast CT of the whole abdomen. This was followed by angiography where AV malformation was seen.[10] The other case had negative abdominal CT angiogram and subsequent colonoscopy showed old blood clots in the entire colon with active bleeding in the appendiceal orifice which was unsuccessfully controlled by epinephrine injection.[15] Both cases underwent appendectomy, and the diagnosis of AV malformation was confirmed through histopathology, similar to our case.

To our knowledge, this was the first reported case in the Philippines and on literature review, the fourth case of an AV malformation of the appendix presenting with lower gastrointestinal bleeding. No further imaging was done in this case since the source of bleeding was identified during colonoscopy.

Our patient underwent laparoscopic appendectomy. As summarized in Table 1, most reported cases of appendiceal bleeding from various causes were managed with appendectomy, either laparoscopic or open. Other treatment strategies that had been reported for appendiceal bleeding included colectomy, clipping, abscess drainage, and therapeutic barium enema.

Conclusion

In summary, we presented a rare case of lower gastrointestinal bleeding caused by AV malformation of the appendix. This case was managed with laparoscopic appendectomy. For patients presenting with hematochezia highly suspicious of colonic etiology with no identifiable source, it is important to examine the appendiceal orifice well during colonoscopy so as not to miss this rare but treatable cause of lower gastrointestinal bleeding.

References

1. Strate LL, Gralnek IM. ACG Clinical Guideline: Management of Patients With Acute Lower Gastrointestinal Bleeding. *Am J Gastroenterol*. 2016;111(4):459-474. doi:10.1038/ajg.2016.41
2. Oakland K, Chadwick G, East JE, et al. Diagnosis and management of acute lower gastrointestinal bleeding: guidelines from the British Society of Gastroenterology. *Gut*. 2019;68(5):776-789. doi:10.1136/gutjnl-2018-317807
3. Imdahl A. Genesis and pathophysiology of lower gastrointestinal bleeding. *Langenbecks Arch Surg*. 2001;386(1):1-7. doi:10.1007/s004230000194
4. Xing XC, Yang JL, Xiao X. Clinical features, treatments and prognosis of appendiceal bleeding: a case series study. BMC Gastroenterol. 2023 Nov 3;23(1):377. doi: 10.1186/s12876-023-03025-6. PMID: 37924017; PMCID: PMC10625278.
5. Aoki T, Hirata Y, Yamada A, Koike K. Initial management for acute lower gastrointestinal bleeding. *World J Gastroenterol*. 2019; 25(1):69-84. doi:10.3748/wjg.v25.i1.69
6. Ghassemi KA, Jensen DM. Lower GI bleeding: epidemiology and management. *Curr Gastroenterol Rep*. 2013;15(7):333. doi:10.1007/s11894-013-0333-5
7. Ma Q, Du JJ. Appendiceal bleeding caused by vascular malformation: A case report. World J Clin Cases. 2024 May 16;12(14):2457-2462. doi: 10.12998/wjcc.v12.i14.2457. PMID: 38765744; PMCID: PMC11099410.
8. Tabcheh A, Salem J, Zodeh K, Ghazale A. Appendiceal Bleeding, A Rare Yet Important Cause of Lower Gastrointestinal Bleed. Eur J Case Rep Intern Med. 2024 Sep 23;11(10):004683. doi: 10.12890/2024_004683. PMID: 39372163; PMCID: PMC11451839.
9. Dung LT, Hung ND, Phuong LTH, et al. Appendiceal hemorrhage: An uncommon cause of lower gastrointestinal tract bleeding and intraluminal contrast extravasation. *Radiol Case Rep*. 2023;18(6):2232-2236. Published 2023 Apr 13. doi:10.1016/j.radcr.2023.03.035
10. Nguyen TA, Van Pham H, Tran TM. Hematochezia due to arteriovenous malformation of the mesoappendix: a rare case report. *J Surg Case Rep*. 2023;2023(4):rjad164. Published 2023 Apr 12. doi:10.1093/jscr/rjad164
11. Zhou SY, Guo MD, Ye XH. Appendiceal bleeding: A case report. *World J Clin Cases*. 2022;10(18):6314-6318. doi:10.12998/wjcc.v10.i18.6314
12. Maeda Y, Saito S, Ohuchi M, Tamaoki Y, Nasu J, Baba H. Appendiceal bleeding in an elderly male: a case report and a review of the literature. *Surg Case Rep*. 2021;7(1):147. Published 2021 Jun 23. doi:10.1186/s40792-021-01234-3
13. Smith EJ, Coventry C, Taylor J, De'ath H, Haque A. A case of endovascular management to gain control of a lower gastrointestinal haemorrhage caused by appendiceal artery bleeding. *J Surg Case Rep*. 2021; 2021(6):rjab204. Published 2021 Jun 10. doi:10.1093/jscr/rjab204
14. Ahn SR, Lee JH. Acute Eosinophilic Appendicitis: A Rare Cause of Lower Gastrointestinal Hemorrhage. *Korean J Gastroenterol*. 2021;78(2):134-137. doi:10.4166/kjg.2021.063
15. Khan F, Lu S, Parajuli D. A Rare Cause of Massive Hematochezia: Arteriovenous Malformation of the Appendix. *ACG Case Rep J*. 2021;8(11):e00679. Published 2021 Nov 18. doi:10.14309/crj.0000000000000679

16. Karatas M, Simsek C, Gunay S, et al. Acute lower gastrointestinal bleeding due to low-grade mucinous neoplasm of appendix. *Acta Chir Belg*. 2022;122(5):357-360. doi:10.1080/00015458.2020.1860397
17. Chen W, Qiu H, Yang X, Zhang J. Appendix Bleeding with Painless Bloody Diarrhea: A Case Report and Literature Review. *Open Med (Wars)*. 2019;14:735-739. Published 2019 Oct 2. doi:10.1515/med-2019-0084
18. Gurien SD, Stright A, Garuthara M, Klein JDS, Rosca M. An iliac-appendiceal fistula causing gastrointestinal bleeding. *J Vasc Surg Cases Innov Tech*. 2019;5(2):107-109. Published 2019 Apr 28. doi:10.1016/j.jvscit.2018.12.008
19. Shen Z, Huang YZ, Ning LM, Gao HC, Wang W. A case of lower digestive tract hemorrhage caused by appendicitis in China. *Int J Surg Case Rep*. 2017;39:34-35. doi:10.1016/j.ijscr.2017.06.038
20. Choi JM, Lee SH, Lee SH, Ahn BK, Baek SU. Hematochezia due to Angiodysplasia of the Appendix. *Ann Coloproctol*. 2016;32(3):117-119. doi:10.3393/ac.2016.32.3.117
21. Magaz Martínez M, Martín López J, De la Revilla Negro J, et al. Appendicular bleeding: an excepcional cause of lower hemorrhage. *Rev Esp Enferm Dig*. 2016;108(7):437-439. doi:10.17235/reed.2015.3828/2015
22. Reynolds JK, Mejia VA. Appendiceal Dieulafoy lesion: an unusual cause of massive lower gastrointestinal bleeding. *Am Surg*. 2015;81(1):E18-E19.
23. Vesa TS, Hosseini-Carroll P, Manas K. Diverticular hemorrhage of the appendix. *Gastroenterol Hepatol (N Y)*. 2014;10(6):394-395.
24. Johnson A, Oger M, Capovilla M. Dieulafoy lesion of the appendix. *Dig Liver Dis*. 2014;46(9):e11. doi:10.1016/j.dld.2014.04.001
25. Konno Y, Fujiya M, Tanaka K, et al. A therapeutic barium enema is a practical option to control bleeding from the appendix. *BMC Gastroenterol*. 2013;13:152. Published 2013 Oct 25. doi:10.1186/1471-230X-13-152
26. Gu MJ, Choi JH, Kim SH. Atypical florid vascular proliferation in appendix: a diagnostic dilemma. *Diagn Pathol*. 2013;8:12. Published 2013 Jan 24. doi:10.1186/1746-1596-8-12
27. Monjero-Ares I, Gegúndez-Gómez C, Rielo-Arias FJ, et al. Iliac-appendiceal fistula. An unusual cause of gastrointestinal bleeding. *Rev Esp Enferm Dig*. 2012;104(7):387-388. doi:10.4321/s1130-01082012000700012
28. Chung IH, Kim KH. A case of successful colonoscopic treatment of acute appendiceal bleeding by endoclips. *J Korean Soc Coloproctol*. 2011;27(6):329-332. doi:10.3393/jksc.2011.27.6.329
29. Chung KS, Gao JP. Massive lower gastrointestinal bleeding from the appendix. *Gut Liver*. 2011;5(2):234-237. doi:10.5009/gnl.2011.5.2.234
30. Chiang CC, Tu CW, Liao CS, Shieh MC, Sung TC. Appendiceal hemorrhage -- an uncommon cause of lower gastrointestinal bleeding. *J Chin Med Assoc*. 2011;74(6):277-279. doi:10.1016/j.jcma.2011.04.009
31. Park I, Kwon CI, Ko KH, Hong SP, Park PW. Colonoscopic clipping as a treatment for appendiceal bleeding. *Gut Liver*. 2010;4(3):411-414. doi:10.5009/gnl.2010.4.3.411
32. Baek SK, Kim YH, Kim SP. Acute lower gastrointestinal bleeding due to appendiceal mucosal erosion. *Surg Laparosc Endosc Percutan Tech*. 2010;20(3):e110-e113.
33. Kim KJ, Moon W, Park MI, Park SJ, Lee SH, Chun BK. Gastrointestinal stromal tumor of appendix incidentally diagnosed by appendiceal hemorrhage. *World J Gastroenterol*. 2007;13(23):3265-3267. doi:10.3748/wjg.v13.i23.3265
34. Lima SE Jr, Speranzini MB, Guiro MP. Doença de Crohn isolada do apêndice cecal como causa de enterorragia [Isolated Crohn's disease of the appendix as a source of enterorrhagia]. *Arq Gastroenterol*. 2004;41(1):60-63. doi:10.1590/s0004-28032004000100012

35. Zitt M, Klaus A, Kirchmayr W, Ammann K, Weiss H, Mühlmann G. Colonoscopically placed hemoclips as treatment for massive appendiceal stump bleeding. *Surg Endosc*. 2003;17(10):1677-1678. doi:10.1007/s00464-003-4219-8
36. Kyokane T, Akita Y, Katayama M, et al. Angiodysplasia of the appendix. *Am J Gastroenterol*. 2001;96(1):242-244. doi:10.1111/j.1572-0241.2001.03385.x
37. Choi DY, Yuh JN, Reid JD. Rectal hemorrhage from ulcerated appendiceal stump nine years after appendectomy. Report of a case. *Dis Colon Rectum*. 1985;28(6):454-456. doi:10.1007/BF02560236
38. Norman DA, Morrison EB, Meyers WM Jr. Massive gastrointestinal hemorrhage from a diverticulum of the appendix. *Dig Dis Sci*. 1980;25(2):145-147. doi:10.1007/BF01308314
39. Mullen JT. Mucocele of the appendix associated with hematochezia. *South Med J*. 1979;72(6):766-767. doi:10.1097/00007611-197906000-00044
40. Milewski PJ. Appendix abscess with intestinal haemorrhage. *Br Med J*. 1977;1(6054):147. doi:10.1136/bmj.1.6054.147