Radiology Case Reports 19 (2024) 315–318

Available online at www.sciencedirect.com

ScienceDirect

journal homepage: www.elsevier.com/locate/radcr

Case Report

A rare cause of rectal bleeding in a 48-year-old lupus patient☆,☆☆

*Oumayma El Eulj, MD**, *Hajar Koulali, MD, Ouiame El Mqaddem, MD, Zahi Ismaili, MD, Ghizlane Kharrasse, MD*

Department of Hepatogastroenterology, Mohammed VI University Hospital Center, Faculty of Medicine and Pharmacy Mohammed I University, Digestive Disease Research Laboratory, Mohammed I University, BP 4806 Oujda University 60049, Oujda 60000, Morocco

ARTICLE INFO

Article history:
Received 11 August 2023
Revised 1 October 2023
Accepted 8 October 2023

Keywords:
Rectal cavernous hemangioma
Gastrointestinal hemangioma
MRI
CT
Endoscopy

ABSTRACT

Cavernous hemangiomas represent a rare benign cause of rectal bleeding. It corresponds to a vascular malformation that can be located anywhere in the gastrointestinal tract. Our paper reports the case of a 48-year-old woman treated for cutaneous lupus who presented to our department with intermittent moderate rectal bleeding. The Hemoglobin level was normal. A colonoscopy showed a congestive nodular red-purple vascular formation. At Computerized tomography (CT)-Scan, the lesion appeared as an irregular thickening of the posterior rectal wall, invading the meso-rectum. Magnetic resonance imaging (MRI) showed a submucosal mass of the rectum containing phleboliths with progressive enhancement in the T2 sequence. Diagnosis of rectal cavernous hemangioma was confirmed. Surgery is the most appropriate treatment for this condition, but since bleeding was not important and had no biological repercussions, multidisciplinary experts meeting decided not to operate on the patient and continue monitoring.

© 2023 The Authors. Published by Elsevier Inc. on behalf of University of Washington. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/)

Introduction

Cavernous hemangiomas belong to a group of benign vascular malformations that can be located anywhere in the gastrointestinal tract. First described in 1839 by Philipps [1], they represent a rare cause of bleeding that often leads to delayed diagnosis and treatment.

Case presentation

A 48-year-old female patient presented to the gastroenterology department as an outpatient for isolated rectal bleeding. She has a history of lymph node tuberculosis at the age of ten and was diagnosed with cutaneous lupus in 2012 and treated with hydroxychloroquine. The patient described intermittent

☆ Acknowledgments: No dedicated source of funding
☆☆ Competing Interests: The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.
* Corresponding author.
E-mail address: eleuljoumayma@gmail.com (O. El Eulj).
https://doi.org/10.1016/j.radcr.2023.10.029
1930-0433/© 2023 The Authors. Published by Elsevier Inc. on behalf of University of Washington. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/)

episodes of rectal bleeding associated with moderate abdominal pain. Her general condition was good. Physical examination was normal. Hemoglobin level, mean corpuscular volume (MCV), mean corpuscular hemoglobin concentration (MCHC), and hematocrit were normal. Colonoscopy showed a congestive formation with serpiginous vessels located in the lower rectum (Fig. 1). CT-Scan found an irregular thickening of the posterior rectal wall infiltrating the meso-rectum (Fig. 2). MRI showed a submucosal mass of the lower rectum measuring 6 cm in length, containing phleboliths. The lesion didn't extend to the muscularis propria nor the sphincter. The mass had a high signal intensity in T2 and diffusion sequences, with a progressive enhancement (Fig. 3). Clinical, endoscopic, and imaging data were in favor of diffuse cavernous rectal hemangioma. Since the bleeding was (mild and intermittent) with no biological - repercussions, a multidisciplinary experts meeting decided to put the patient under clinical and imaging monitoring.

Discussion

Gastrointestinal hemangiomas are a group of benign vascular malformations; their incidence is estimated at 0.3% [2]. They correspond to a network of vascular structures located in the intestinal wall [3]. They are classified as capillary, cavernous, arteriovenous, and other mixed types. About 80% of rectosigmoid hemangiomas are cavernous [4]. The suspected physiopathological mechanism is an abnormal development of mesodermal tissue that occurs during prenatal period [5]. Unregulated angiogenesis is also hypothesized as a possible explanation for hemangiomas, and the association with systemic lupus erythematosus (SLE), as suggested by Berzigotti et al. [6], would be caused by an imbalance between angiostatic and angiogenic factors usually increased in active SLE patients. At the diagnosis, most of the patients are young adults who present with chronic and recurrent rectal bleeding,

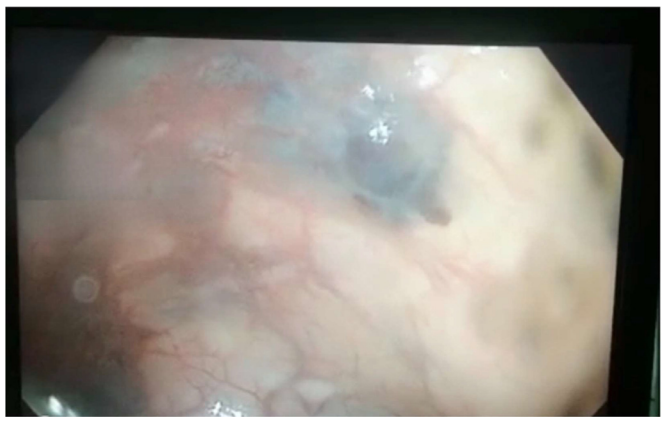

Fig. 1 – Colonoscopy showed a rectal (low rectum) congestive formation with serpiginous vessels.

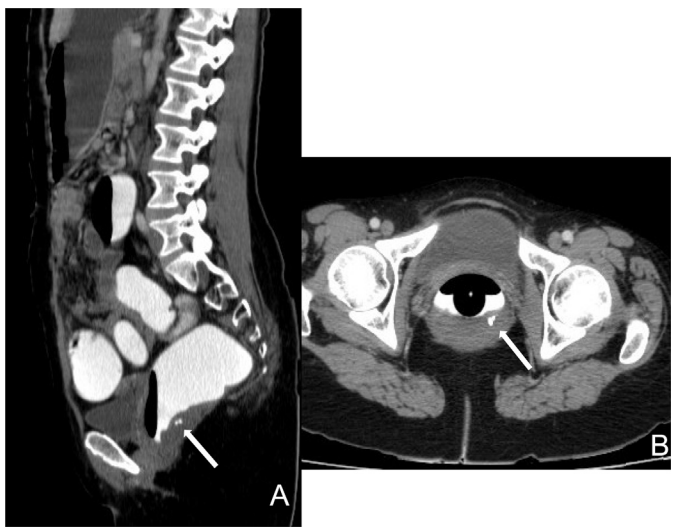

Fig. 2 – CT-Scan: Sagittal (A) and Axial (B) sections with gastrografin opacification showed a posterior rectal mass containing phleboliths.

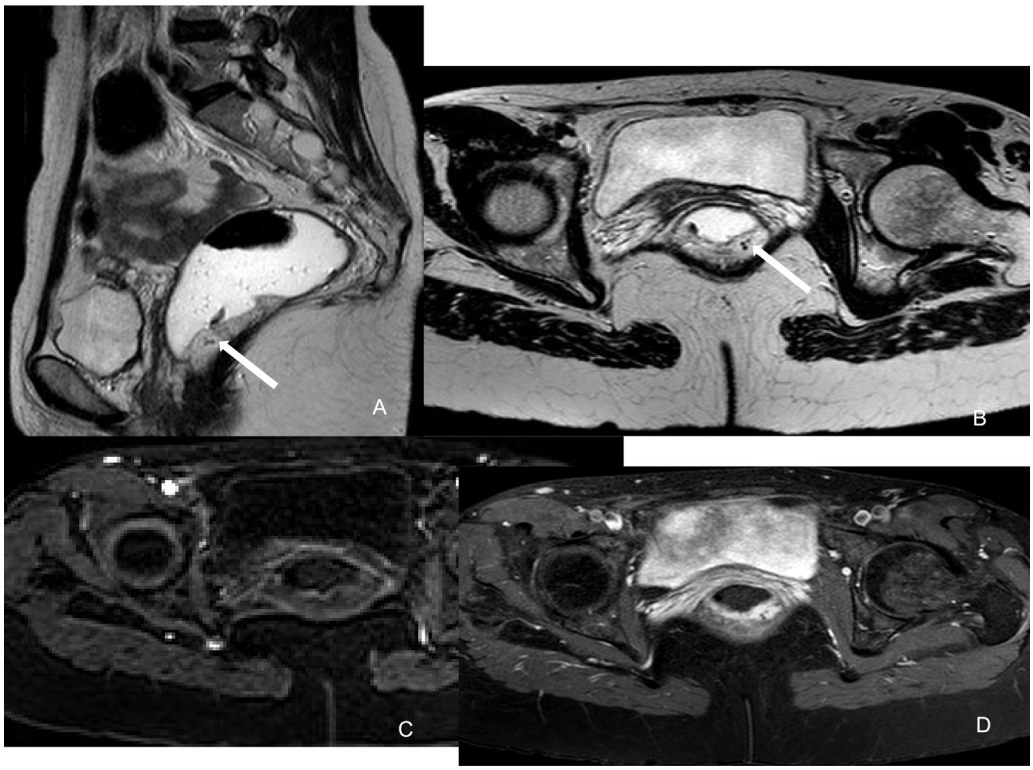

Fig. 3 – MRI Sagittal T2 (A), Axial T2 (B), injected axial T1 in arterial (C) and tardive (D) phases showed a mass of posterior rectal wall with regular contours, with high-signal in T2, containing phleboliths (arrows), with progressive enhancement and perirectal vascular turgitidy.

ongoing since childhood. Hemorrhage can be massive and life-threatening. Isolated anemia can be the only sign when bleeding is occult. Other symptoms, such as abdominal or pelvic pain, obstruction, or perforation are due to compression [7]. Due to the chronicity and rarity of the disease, the average delay of diagnosis is estimated at 19 years in the literature [5]. Reported symptoms are often misdiagnosed as ulcerative colitis, Crohn's disease, malignant tumors, or internal hemorrhoids, leading to at least one misled surgery before the diagnosis in 80% of patients [2]. Endoscopy often describes a soft and compressible bluish or deep red submucosal lesion, with dilated and engorged veins in the rectal wall as was seen in our patient [7]. In some cases, rectal ischemia can be caused by the obstruction of supply vessels, leading to chronic inflammatory changes such as mucosal edema and ulcerations, explaining the misdiagnose of proctitis [8]. Biopsy should not be performed because of a high risk of an uncontrollable bleeding. CT and MRI play a major role in diagnosis accuracy. They can define the extension of the lesion and its relation to other structures [9]. CT imaging allows visualization of calcifications (phleboliths), that are characteristic in cavernous hemangiomas [5], as observed in our patient. In their paper, Yesilkaya et al. described MRI features as follows: intermediate signal intensity on T1-weighted images and high signal intensity on T2-weighted ones. Thrombosed vessels appear as tubular structures with high signal intensity. Signal voids can represent calcified regions of the lesion and blood vessels [3]. Together with endoscopy, CT, and MRI have a complementary role in diagnosis assessment. Therapeutic approach is determined by the intensity and impact of symptoms, but also the location and size of the lesion. Sclerotherapy, argon fulguration, or cryotherapy have been used and are only suitable for well-defined small lesions. In case of acute bleeding, embolization can be useful, but not as a definitive treatment [5]. Complete surgical resection is the preferred therapy for bleeding control, and consists of an anterior rectal resection and colo-anal anastomosis, with sphincter conservation [2]. Abdominoperineal resection can be required if the lesion extends into the perineum, gluteal regions, or anal canal [7]. Although considered a benign condition, cavernous hemangioma can still be life-threatening in case of massive bleeding.

Conclusion

Cavernous hemangioma is a rare cause of rectal bleeding and requires a good knowledge of the condition to avoid misdiagnoses and inappropriate therapies. Endoscopy and imaging are the pillars of the diagnosis. When suspected, biopsy should be avoided due to a high bleeding risk. Surgery remains the most effective treatment to control the possible bleeding that can be massive and life-threatening.

Author contributions

El Eulj O collected the patients' data, designed the report, and wrote the paper; Koulali H, El Mqaddem O, and Ismaili Z participated in the multidisciplinary teams meeting; Kharrasse G performed endoscopy and supervised the writing and editing of the report; All authors read and approved the manuscript.

Patient consent

Informed consent was obtained from the patient for the publication of their case.

REFERENCES

[1] Andrade P, Lopes S, Macedo G. Diffuse cavernous hemangioma of the rectum: case report and literature review. Int J Colorectal Dis 2015;30(9):1289–90. doi:10.1007/s00384-015-2329-0.
[2] Hermosa AR, Zorrilla-Ortuzar J, Valle-Hernández ED. Diffuse cavernous hemangioma of the rectum. Cir Cir 2021;89(6):818–21. doi:10.24875/CIRU.20000746.
[3] Yeşılkaya Y, Hızal M, Akata D. Infiltrating rectal cavernous hemangioma mimicking rectal tumor. Turk J Gastroenterol 2012;23(2):190–1. doi:10.4318/tjg.2012.0336.
[4] González MM, Torcuato ÁP, Jimenez MLM, Sebastián IA, Porras JLM, Egea GS, et al. Recurrent rectal bleeding in a teenager caused by a cavernous haemangioma of the sigmoid colon. Lancet 2022;400(10369):2249. doi:10.1016/S0140-6736(22)02035-9.
[5] Stojčev Z, Maliszewski D, Pawłowska-Stojčev I, Kasprzyk T, Jaśkiewicz J. Diffuse cavernous hemangioma of the rectum (DCHR)–diagnosis and treatment–case report and review of available literature. Pol Przegl Chir 2013;85(4):216–18. doi:10.2478/pjs-2013-0033.
[6] Berzigotti A, Frigato M, Manfredini E, Pierpaoli L, Mulè R, Tiani C, et al. Liver hemangioma and vascular liver diseases in patients with systemic lupus erythematosus. World J Gastroenterol 2011;17(40):4503–8. doi:10.3748/wjg.v17.i40.4503.
[7] Sylla P, Deutsch G, Luo J, Recavarren C, Kim S, Heimann TM, et al. Cavernous, arteriovenous, and mixed hemangioma-lymphangioma of the rectosigmoid: rare causes of rectal bleeding: case series and review of the literature. Int J Colorectal Dis 2008;23(7):653–8. doi:10.1007/s00384-008-0466-4.
[8] Chatu S, Kumar D, Du Parcq J, Vlahos I, Pollok R. A rare cause of rectal bleeding masquerading as proctitis. J Crohns Colitis 2013;7(3):e99–102. doi:10.1016/j.crohns.2012.05.022.
[9] Hervías D, Turrión JP, Herrera M, Navajas León J, Pajares Villarroya R, Manceñido N, et al. Diffuse cavernous hemangioma of the rectum: an atypical cause of rectal bleeding. Rev Esp Enferm Dig 2004;96(5):346–52 English, Spanish. doi:10.4321/s1130-01082004000500008.