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Congenital Colon Arteriovenous Malformation Misdiagnose in a 25-Year-Old Woman

Saeed Derakhshani ^{1,2}; Mehran Babaei ²; Mojgan Forootan ¹; Nakisa Maghsoodi ¹; Seyed Ali Majdzadeh ^{2,*}

¹Research Center for Gastroenterology and Liver Diseases, Shahid Beheshti University of Medical Sciences, Tehran, IR Iran ²Parsian Hospital, Tehran, IR Iran

*Corresponding author: Seyed Ali Majdzadeh, Parsian Hospital, Tehran, IR Iran. Tel: +98-9359356667, Fax: +98-2122938262, E-mail: majdzadeh@outlook.com

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Dear Editor

Congenital arteriovenous malformations (AVMs) are uncommon lesions. Any developmental arrest or misdirection, during the formation of the capillary network can lead to communicating channel formation, between mature arteries and veins. These channels are responsible for the development of additional branches, due to the overgrowth of vascular elements. Aging of the AVM component elements leads to increase of its size, which may lead to symptoms (1). Pelvic AVMs are rare and are recognized as one of the rare causes of rectal bleeding (2). While many of the conventional diagnostic studies are not very helpful for detecting GI system AVMs if the patient has any signs or symptoms suggesting this diagnosis, other diagnostic tests like CT scan and color flow Doppler sonography can be helpful (3-5).

The most important symptom in ulcerative colitis (UC), especially at the early stages, is rectal bleeding, with or without mucus (6). Colon AVMs are not among the most prevalent rectal bleeding causes, but should be considered as one of the differential diagnoses (7).

Our patient was a 25-year-old married woman with a 6-year history of episodic rectal bleeding. The patient did not experience any other associated symptoms includ-

ing diarrhea, constipation or abdominal pain. She experiences several periods of severe anemia, due to rectal bleeding during 6 years. She reported infertility despite trying for two years. Her grandmother, aunt and cousin were known cases of UC. Two years prior to admission, during a bleeding episode, the patient underwent a diagnostic colonoscopy and biopsy. The pathology report mentioned proctosigmoiditis, a common finding in UC. Based on the family history and the pathology report, the patient was diagnosed as a new case of UC treated with mesalazine and prednisone without any improvements in her condition, including rectal bleeding episodes pattern, severity and volume. She needed several blood transfusion sessions, during her medical treatment course, due to her low hemoglobin levels. Ultimately she was referred to the surgical team as a candidate for the ileal pouch-anal anastomosis (IPAA) surgical treatment of UC. As a standard procedure a second opinion on pathology samples was requested before IPAA. The second pathologist's opinion contradicted the preliminary report, described the slides as a clear case of AVM. To decide the best course of treatment, the patient went under her second colonoscopy. This time venous engorgement was observed in distal sigmoid, extending to the anal canal (Figure 1).

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Figure 1. Diffused Angioectasias, With Red and Blue Patterns Seen in Rectum, Sigmoid and Descending Colon

Due to the vast extension of the AVM and as a diagnostic and therapeutic procedure, the patient went under a surgical exploration. During the laparotomy the AVMS were detected in pelvis, around the uterus, distal sigmoid and rectum. Low anterior and sigmoid resection, colonic J-pouch-anal anastomosis and diverting ileostomy were performed during the laparotomy. After 45 days, the ileostomy was removed. Patient experienced mild rectal bleeding and abdominal pain, following the surgical treatment. At the 6 month follow-up, the patient was completely symptom free, with a normal bowel habit.

Congenital AVMs rarely cause rectal bleeding (7). While bloody diarrhea is the hallmark of UC, over time the patients experience more inflammatory symptoms, including urgency or tenesmus, abdominal pain, fever, malaise and weight loss (6). In this case, massive rectal bleeding was the only symptom. Unfortunately, the patient was misdiagnosed preliminary, based on her family history and an inaccurate pathology result and took two years of unnecessary medical UC treatment. It shows the importance of congenital AVMs, as the differential diagnosis of rectal bleeding. Detection of colon AVMs might need a combination of three different diagnostic approaches; angiography, colonoscopy and even surgical exploration, but the definite diagnosis method is the histological demonstration of the AVM (2). Although other treatments, like selective mesenteric angiography (SMA) are available for limited AVMs of colon, but with serious side effects, when the bleeding volume is massive (8). Additionally, in this case anus was involved .Its dual blood supply makes the angiographic approach difficult. Therefore, surgical exploration was the most appropriate approach, due to the size of the AVM, history of several massive bleedings and involvement of the anal canal. The uterine and pelvic AVMs are not easily diagnosed if asymptomatic (9). In this scenario, the pelvic and uterine AVMs were detected during the surgery, while they could remain undetected.

Authors' Contributions

Dr. Saeed Derakhshani performed the surgery and supervised the project. Dr. Mehran Babaei and Dr.Mojgan Forootan were the gastroenterologists involved in patients diagnosis and treatment. Dr. Nakisa Maghsoodi gathered data. Dr. Seyed Ali Majdzadeh is the Corresponding Author and gathered data and wrote the article.

References

- Gomes MM, Bernatz PE. Arteriovenous fistulas: a review and tenyear experience at the Mayo Clinic. *Mayo Clin Proc.* 1970;45(2):81– 102.
- Hagihara PF, Chuang VP, Griffen WO. Arteriovenous malformations of the colon. Am J Surg. 1977;133(6):681–7.
- Fakhri A, Fishman EK, Mitchell SE, Siegelman SS, White RI. The role of CT in the management of pelvic arteriovenous malformations. *Cardiovasc Intervent Radiol.* 1987;10(2):96–9.
- Abu Musa A, Hata T, Hata K, Kitao M. Pelvic arteriovenous malformation diagnosed by color flow Doppler imaging. AJR Am J Roentgenol. 1989;152(6):1311-2.
- Jain KA, Jeffrey RB, Jr, Sommer FG. Gynecologic vascular abnormalities: diagnosis with Doppler US. Radiology. 1991;178(2):549–51.
- Danese S, Fiocchi C. Ulcerative colitis. N Engl J Med. 2011;365(18):1713-25.
- Foutch PG, Rex DK, Lieberman DA. Prevalence and natural history of colonic angiodysplasia among healthy asymptomatic people. *Am J Gastroenterol.* 1995;90(4):564–7.
- Kontopodis N, Spiridakis K, Grigoraki M, Panagiotakis G, Papadakis T, Kokkinakis T, et al. Colon bleeding from both arteriovenous malformation and inflammatory bowel disease. Case report. G Chir. 2011;32(10):417-20.
- Kushtagi P, Rao K, Ramkumar V. Congenital pelvic arteriovenous malformation with massive vaginal bleeding. Int J Gynaecol Obstet. 1996;52(1):71-2.