

CASE REPORT

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Revisiting imaging features of rectosigmoid vascular malformation with emphasis on multiparametric MRI: a case report

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Abstract

Background: Gastrointestinal vascular malformations are rare benign vascular neoplasms of the gastrointestinal tract, with the rectosigmoid region being the most frequently involved site. Patients often manifest with recurrent, intermittent rectal bleeding, which might occasionally be life-threatening.

Case presentation: A 39-year-old man with a history of hemorrhoid operations twice was presented to our gastroenterology department with blood in the stool and abdominal pain. After the colonoscopy, multiparametric MRI, and CT examinations, robotic low anterior resection was performed with the diagnosis of rectosigmoid venous malformation. The histopathological examination confirmed the diagnosis.

Conclusion: Colonoscopy is the preferred method in diagnosing rectosigmoid vascular malformation, but wrong and delayed diagnoses are common. Thus, imaging modalities might add to colonoscopy in equivocal cases.

Keywords: Magnetic resonance imaging, Rectum, Vascular anomalies, Colonoscopy, Hemangioma

Background

Rectal bleeding is the most common indication of colonoscopy. Though colon cancer is one of the common causes of rectal bleeding, underlying vascular anomalies should be considered in the differential diagnosis [1]. According to the 2018 International Society for the Study of Vascular Anomalies (ISSVA) classification, vascular anomalies are divided into vascular tumors and malformations [2]. Patients with rectosigmoid venous malformation often present recurrent, intermittent rectal bleeding, which might be slow or massive and life-threatening.

Colonoscopy is the reference modality in diagnosing rectosigmoid venous malformations, yet misdiagnosis is not uncommon, leading to inappropriate or delayed

treatments [3]. Magnetic resonance imaging (MRI) and computed tomography (CT) might contribute to colonoscopy in challenging cases, sparing patients from unnecessary surgical interventions, and allowing them to receive the most suitable treatment without delay. CT might be of value by showing diffuse wall thickening and calcifications due to phleboliths, while MRI might show T2W hyperintense diffuse wall thickening accompanied by enlarged serpentine vascular structures in the surrounding mesentery and progressive contrast enhancement [3].

This report presents a patient with a rectosigmoid vascular malformation and a history of unnecessary hemorrhoid operations twice. In our patient, the correct diagnosis and decision-making can only be made based on the findings of the multiparametric MRI.

Case presentation

A 39-year-old man who had a history of hemorrhoid operations twice was admitted to our gastroenterology department with blood in the stool and abdominal pain

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lasting three months. The physical examination reveals venous enlargement in the anal canal. The laboratory results showed low hemoglobin (9,9 g/dl) and serum

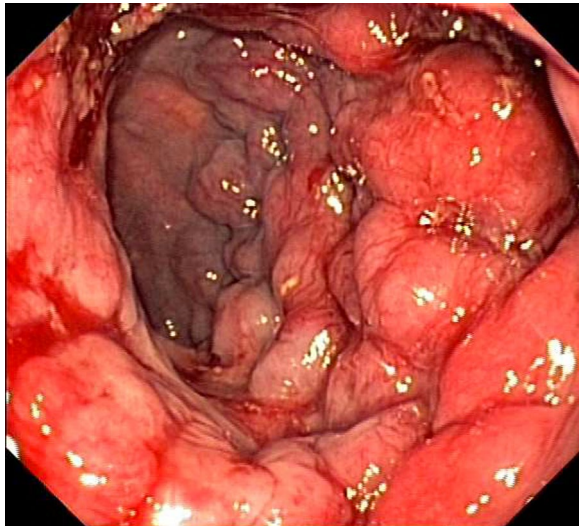


Fig. 1 The colonoscopy image shows dilated submucosal nodular lesions accompanied by edematous mucosal alterations

ferritin (12 ng/ml). The patient underwent a colonoscopy that revealed the nodular submucosal masses are accompanied by inflammatory and edematous alterations in the rectosigmoid wall (Fig. 1).

The patient was referred to MRI for further investigations. A comprehensive MRI including conventional sequences, T1W, T2W imaging, dynamic contrast-enhanced (DCE) T1W perfusion imaging using 7 mL of intravenous contrast-medium (gadobutrol, Gadovist, Bayer, Germany), and diffusion-weighted imaging (DWI) was carried out on a 3 T MRI scanner (GE Signa Premier; GE Healthcare; Chicago, Illinois, U.S.A.). The T2W images revealed diffuse circumferential hyperintense mass encompassing millimetric hypointense foci involving the entire rectosigmoid colon (Fig. 2). The mass did not exhibit prominent early enhancement on the DCE and had low Ktrans values on perfusion. The hypointense foci identified on T2W images suggested multiple calcifications related to phleboliths, which were confirmed on a subsequent non-contrast low-dose CT (Siemens Somatom Force; Siemens Healthcare AG; Erlangen, Germany) (Fig. 3).

The lesion did not exhibit diffusion restriction and showed prominent homogenous contrast enhancement

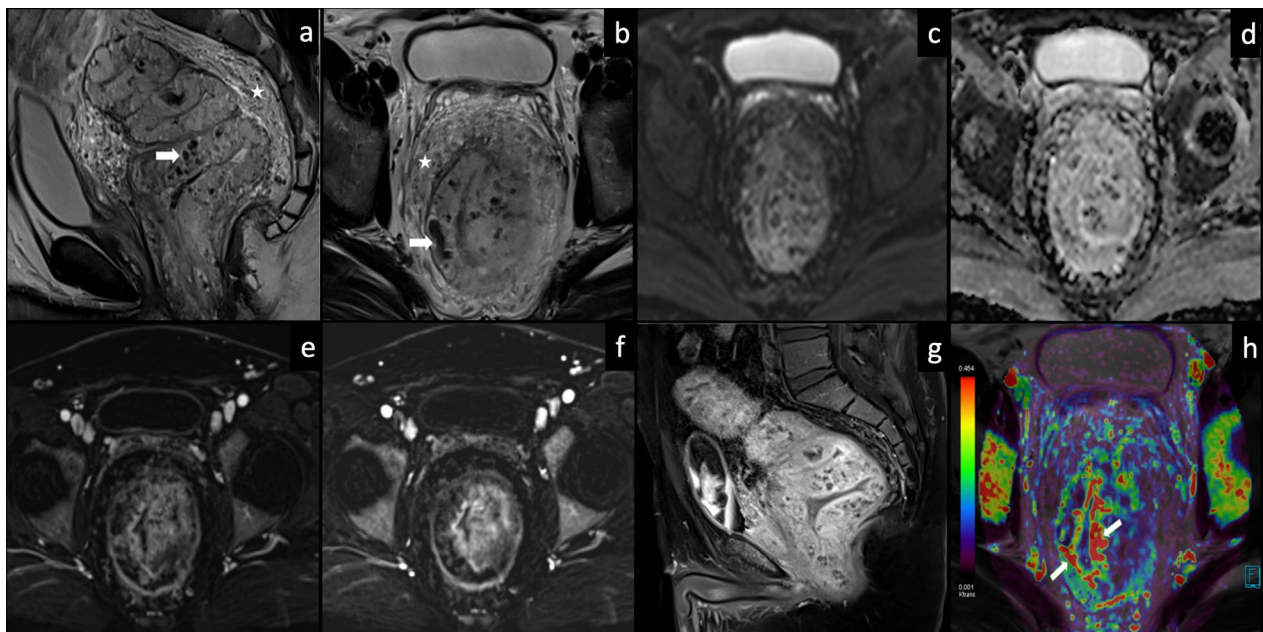


Fig. 2 The sagittal (a) and axial (b) T2W MR images show diffuse thickening in the wall of the rectosigmoid colon characterized by increased signal intensity. The hypointense serpiginous areas represent dilated tortuous venous structures (thick arrows), and multiple round hypointense foci represent phleboliths. The mesorectal fascia has increased heterogenous signal intensity (asterisk). The lesion does not show diffusion-restriction on DWI (c) and ADC map (d). The lesion does not show prominent early or late contrast enhancement on the axial dynamic contrast-enhanced T1W images (e, f). However, the sagittal delayed contrast enhancement image (g) reveals that the lesion has heterogenous diffuse contrast enhancement (g). Overall, the lesion shows decreased perfusion (e) interspersed with highly perfused areas consistent with the dilated venous structures (arrows) (h)

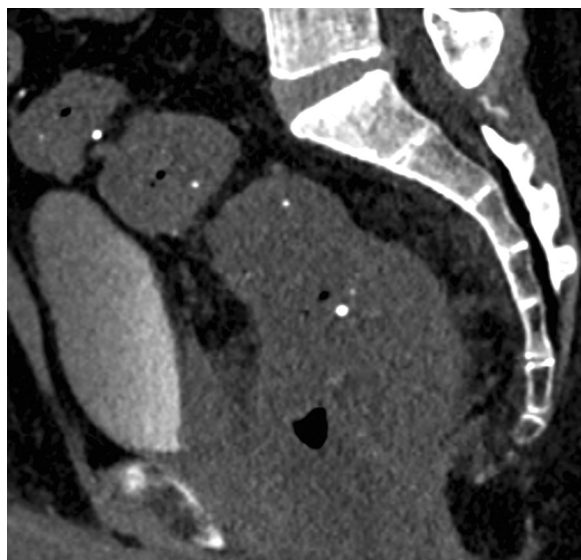


Fig. 3 The non-contrast sagittal CT image shows multiple round foci of calcification consistent with phleboliths

in the delayed phase (Fig. 2). The combination of clinical findings, imaging studies, and colonoscopy was interpreted as a diffuse vascular malformation of the rectosigmoid colon. The patient underwent low anterior resection due to the ongoing bleeding, and the histopathological examination showed diffuse vascular malformation with arterial and venous structures (Fig. 4).

Discussion

This report demonstrated the added value of cross-sectional radiological imaging methods to colonoscopy in diagnosing diffuse rectosigmoid vascular malformation. Though colonoscopy is the reference modality in assessing diffuse rectosigmoid vascular malformation, the disease might be misdiagnosed as the diagnostic capacity of colonoscopy is limited beyond the colonic lumen and mucosa. On the other hand, MR and CT examinations can demonstrate the entire wall and extraluminal extensions, which might be necessary for diagnosing rectosigmoid vascular malformation. Thus, combining colonoscopy with MRI and CT scans might allow a holistic assessment of rectosigmoid vascular malformations [4].

The patient underwent unnecessary surgical interventions after being incorrectly diagnosed with hemorrhoids on colonoscopy. Indeed, the correct diagnosis and successive appropriate treatment were only made possible after MRI and CT were carried out, substantiating the importance of combining colonoscopy with imaging methods in rectosigmoid vascular malformations.

Several accounts of rectosigmoid vascular malformations exist in the literature [5–8]. Most of these papers

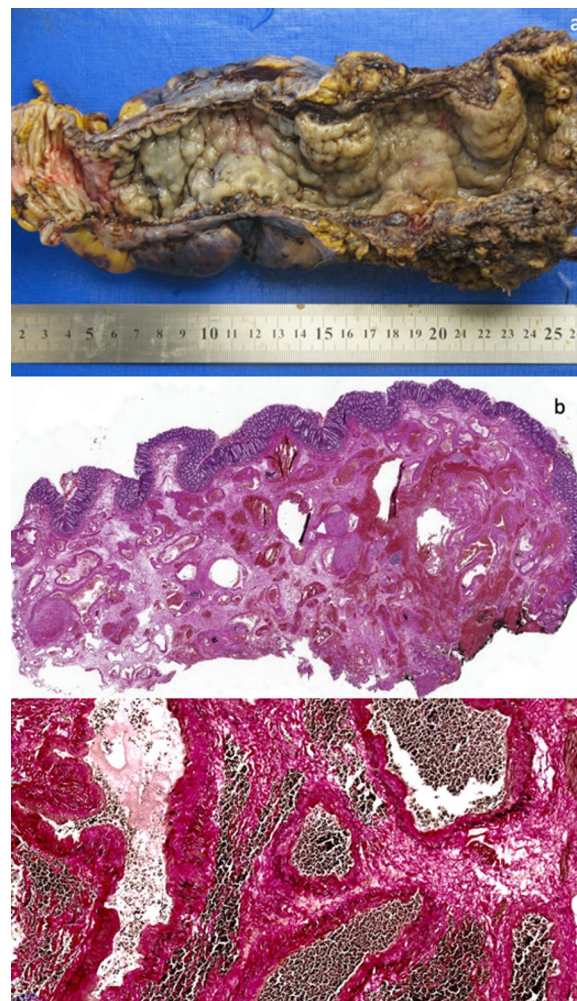


Fig. 4 The macroscopic (a) and microscopic (b, c) specimen images. Macroscopically, the appearance of the bowel mucosa resembles extensive varices. The bluish submucosal tortuous dilation of blood vessels occupies the rectum and distal sigmoid colon. The microscopic examination shows numerous disorganized, dilated, and irregularly thickened abnormal blood vessels. There is a combination of new and organized-calcified thrombi. The vessels have a conspicuous muscular layer formed by the arterial type and venous type elastic fibers

were published before the 2010s; thus, the imaging findings were inevitably restricted to that era's technology. Nonetheless, some of these reports were published last decade [4, 9–11]. In their large sample of children with vascular malformation of the colorectum, Peng et al. showed the yields of ultrasonography in the diagnosis [4]. Abeysekera et al. demonstrated CT and MRI findings of a patient with an incidentally detected rectal vascular malformation in an older man. In their report, the CT scan depicted wall calcifications, and the MRI scan showed diffuse T2W hyperintense wall thickening accompanied

by early nodular wall enhancement on the post-contrast images.

The imaging findings provided by Abeysekera et al. bear a close resemblance to the findings in our case, except for the lack of early nodular contrast enhancement [9]. Furthermore, their patient was asymptomatic and did not undergo an operation, while the patient presented in this report had typical symptoms of diffuse rectosigmoid vascular malformation and clinical history and underwent an operation. Additionally, we presented T1W perfusion and DWI MR imaging features of the disease as well as the images of the histopathological specimen.

DWI and DCE MR perfusion imaging are non-invasive surrogate markers showing the physiological and biological alterations at the cellular level [12]. DWI measures the extracellular random Brownian motion of water, and diffusion-restriction can be a sign of malignancy or inflammation [13]. DCE MR perfusion allows measuring the contrast-uptake of the tissues [12]. Ktrans measured on the perfusion maps is a surrogate marker of capillary permeability, and increased Ktrans is associated with malignancy and inflammation [13]. In our patient, low Ktrans values and lack of diffusion-restriction facilitated excluding diseases of inflammatory origin and cancer and improved the confidence in diagnosing the patient with vascular malformation. Additionally, non-contrast CT was valuable as it showed phleboliths in our case.

Though we did not use conventional angiography in this patient, we acknowledge that mesenteric angiography may contribute to the diagnosis by showing increased blood flow or contrast extravasation in the rectosigmoid region, especially in the delayed venous phases [14]. Furthermore, angiographic embolization might represent an alternative treatment option in selected patient groups (4, 15). Nevertheless, the best treatment method in patients with gastrointestinal vascular malformation should be decided in multidisciplinary meetings.

Conclusions

Though colonoscopy is the reference method of choice and typically could solely diagnose rectosigmoid vascular malformation, misdiagnosing the disease as a hemorrhoid is not uncommon. The disease is accompanied by dilated vessels, occasionally occluded by multiple thrombi formation, leading to inflammatory changes in the rectosigmoid colon. Hence, if the colonoscopy is performed during this phase, it might lead to misdiagnosis of rectitis or cancer. Indeed, a conclusive diagnosis was not possible in the present case on the colonoscopy. Thus, imaging methods, MRI, and CT might be essential in excluding potential pitfalls and confirming the diagnosis.

Abbreviations

DCE: Dynamic contrast-enhanced; DWI: Diffusion-weighted imaging; ISSVA: International Society for the Study of Vascular Anomalies; MRI: Magnetic resonance imaging; CT: Computed tomography.

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Author contributions

SE-writing; DA-editing; BÇ-Data curation and critically reviewed; SG-Data curation and critically reviewed; TK-Data curation and critically reviewed; EK-Data curation and critically reviewed and conceptualization. All authors read and approved the final manuscript.

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Availability of data and materials

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Declarations

Ethics approval and consent to participate

The ethics committee approved this case report and waiver denied for informed consent.

Consent for publication

All authors approved this publication.

Competing interests

The authors declare that they have no competing interests.

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