Case Report

An Incidental Large Rectal Polyp with Idiopathic Small Arteriovenous Dysplasia in Colonic Submucosa: A Case Report

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Rectal polyps with idiopathic small arteriovenous dysplasia are rare. The mesenteric vascular diseases were recently classified as two vascular diseases: fibromuscular dysplasia (FMD) of artery and mesenteric arteriovenous dysplasia/vasculopathy (MAVD/V). FMD usually involves medium size mesenteric arteries in younger individuals. In contrast, MAVD/V tends to affect multiple small mesenteric arteries and veins without vascular lesions in other organs. We reported that a 45-year-old male with a large rectal polyp for routine colorectal screening. Microscopic examination shows chronic ischemic changes and multiple small arteries, veins and capillaries with intimal and medial hyperplasia and focal occlusion, mimicking mesenteric arteriovenous dysplasia and named it as submucosal arteriovenous dysplasia.
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Key Words: *mesenteric arteriovenous dysplasia/vasculopathy, rectal polyp; submucosal arteriovenous dysplasia, MSVD/V*

INTRODUCTION

Mesenteric arteriovenous dysplasia/vasculopathy (MSVD/V) is a rare vascular disorder characterized by non-inflammatory, non-atherosclerotic abnormalities in the mesenteric arterial and venous systems.[1-4] It represents a distinct entity separate from fibromuscular dysplasia (FMD) and has gained recognition as an important cause of mesenteric ischemia.[2,5] Unlike FMD, which primarily affects the renal and internal carotid arteries, MSVD/V involves the mesenteric arterial bed, colonic wall including muscularis propria and submucosa.[2,5] This condition is characterized by concentric/eccentric smooth muscle collarettes surrounding the tunica media of both the artery and the vein in multiple foci, along with varying degrees of intimal and medial hyperplasia and adventitial fibrosis.[2] Importantly, MSVD/V exhibits a lack of inflammation or thrombi, further distinguishing it from other vascular pathologies. The clinical presentation and management of MSVD/V remain poorly understood due to its rarity and limited case reports. The definite diagnosis must be sampled from colorectal resection and the diagnosis from biopsy or mucosal resection is incredibly challenging. Here we report a case of MSVD/V-like disease that was presented as an incidentally found rectal polyp from endoscopic mucosal resection.

CASE REPORT

A 45-year-old male with no significant past medical history was initially evaluated in the outpatient setting for hemorrhoid management. Given his age, he was recommended to undergo routine screening colonoscopy which revealed hemorrhoids and a large edematous rectal lesion, consistent with rectal adenoma. The patient remained asymptomatic and there was no known family history of any GI malignancies. Lab values including complete blood count, coagulation profile, and complete metabolic panel were within normal limits. Due to the size of the lesion, he was referred to the interventional endoscopy team for repeat colonoscopy with hybrid endoscopic submucosal dissection.

During the procedure, a 6.0 cm sessile polyp was again visualized in the rectum, with the distal half of the lesion appearing deeply scarred and fibrosis (**Figure 1**). An attempt was made to endoscopically dissect the lesion from the underlying deep layers with the electrocautery knife, however, the wall layers would not separate well due to the significant

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fibrosis. The decision was then made to perform modified endoscopic mucosal resection and 85% of the lesion was resected. The remaining portion was unable to be removed, and there appeared to be deeper invasion of the lesion into the muscular layer. The patient was discharged home with a plan to repeat colonoscopy in 3 months with EndoRotor resection of remaining polyp tissue if the lesion was benign or refer to colorectal surgeon if there was advanced pathology.

Gross examination of the specimen revealed seven nodular fragments of yellow-tan firm tissue, ranging in size from 0.8-1.3 cm, from the partially resected polypoid lesion. Microscopic examination found that the colonic mucosa and submucosa showed chronic ischemic changes including effacement of superficial epithelium, capillary congestion, hyalinized and fibrosis (**Figure 2A**). Further microscopic examination showed multiple small arteries, veins and capillaries in colonic mucosa and submucosa, with intimal and medial hyperplasia and focal occlusion (**Figure 2B & 2C**). CD34 immunostain highlights the dense dilated small capillaries in the mucosa (**Figure 2D**); PAS-D special stain highlights intimal and medial hyperplasia with collagen-like material (**Figure 2E**); and Trichrome special stain highlights fibrosis around bundles of small arteries and veins. Based on the H&E and other special stains, the diagnosis of submucosal arteriovenous dysplasia is favored. The mesenteric vessels cannot be evaluated due to endoscopic mucosal resection samples. However, based on endoscopic examination of the deeper layer involved by the polyp, it is indicative of possible mesenteric vessel change.

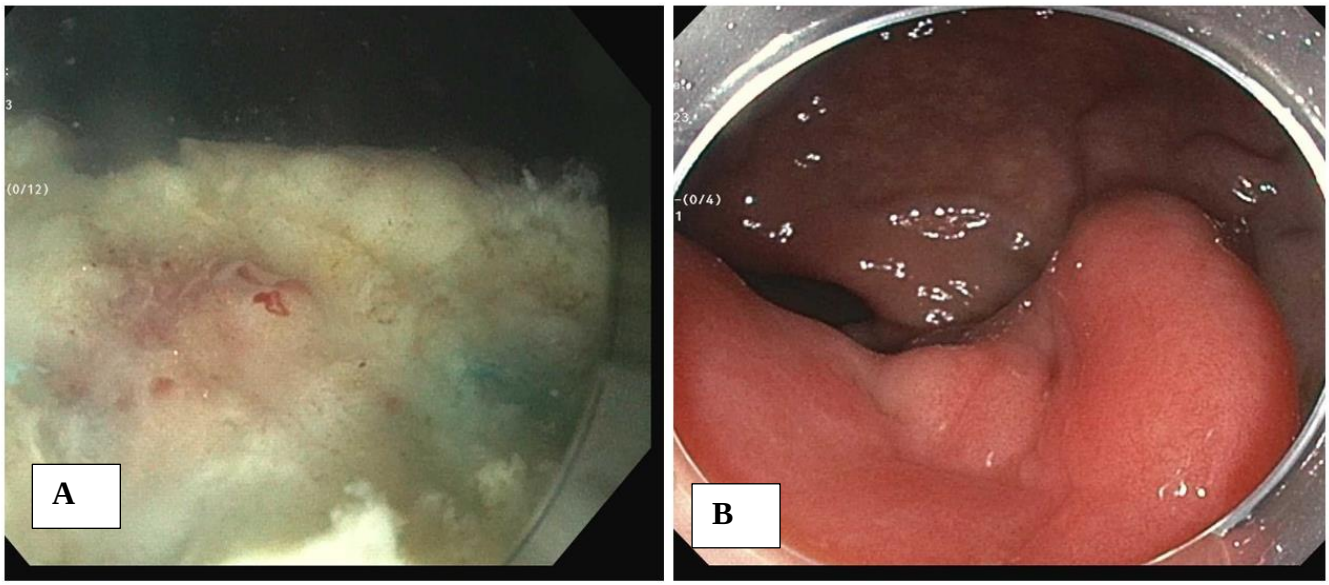

Figure 1. Endoscopic examination shows a 6.0 cm sessile edematous polyp in the rectum. (**A**) The sessile polyp with top surface view; (**B**) The sessile polyp with side view.

DISCUSSION

We report a case of submucosal arteriovenous dysplasia that was presented as an incidental finding of a large rectal polyp in an asymptomatic patient during a routine screening colonoscopy. Microscopic examination showed multiple small arteries and veins with intimal and medial hyperplasia as well as extensive intra-mucosal capillary proliferation in colonic mucosal with chronic ischemic change. The morphological features mimic the MAVD/V.

The terminology regarding mesenteric vascular diseases has only been recently clarified.[2] However, the diagnosis of MSVD/V was made based on the histopathological features described below by Patil et al. in their recent study. These features include: (1) concentric/eccentric smooth muscle collarette around the tunica media of both the artery and the vein in ≥ 2 foci, (2) varying degrees of intimal and medial hyperplasia and adventitial fibrosis, and (3) lack of inflammation or thrombi. Notably, MAVD/V typically involves multiple small mesenteric arteries and veins without associated vascular lesions in other organs. In our case, the small vessels show intimal and medial hyperplasia and adventitial fibrosis without inflammation or thrombi inside of vessels, which are mimic of number 2 and 3 features. However, concentric smooth muscle around the tunica medial cannot be evaluated due to the smaller vessels inside of mucosa and submucosa. However, the compensatory capillary proliferation, ischemic mucosa and deeper layer infiltration indicate other features similar to MSVD/V. Since mesenteric vessels cannot be evaluated by endoscopic mucosal resection samples, we proposed a similar diagnosis terminology for this small vessel's dysplasia as a submucosal arteriovenous dysplasia.

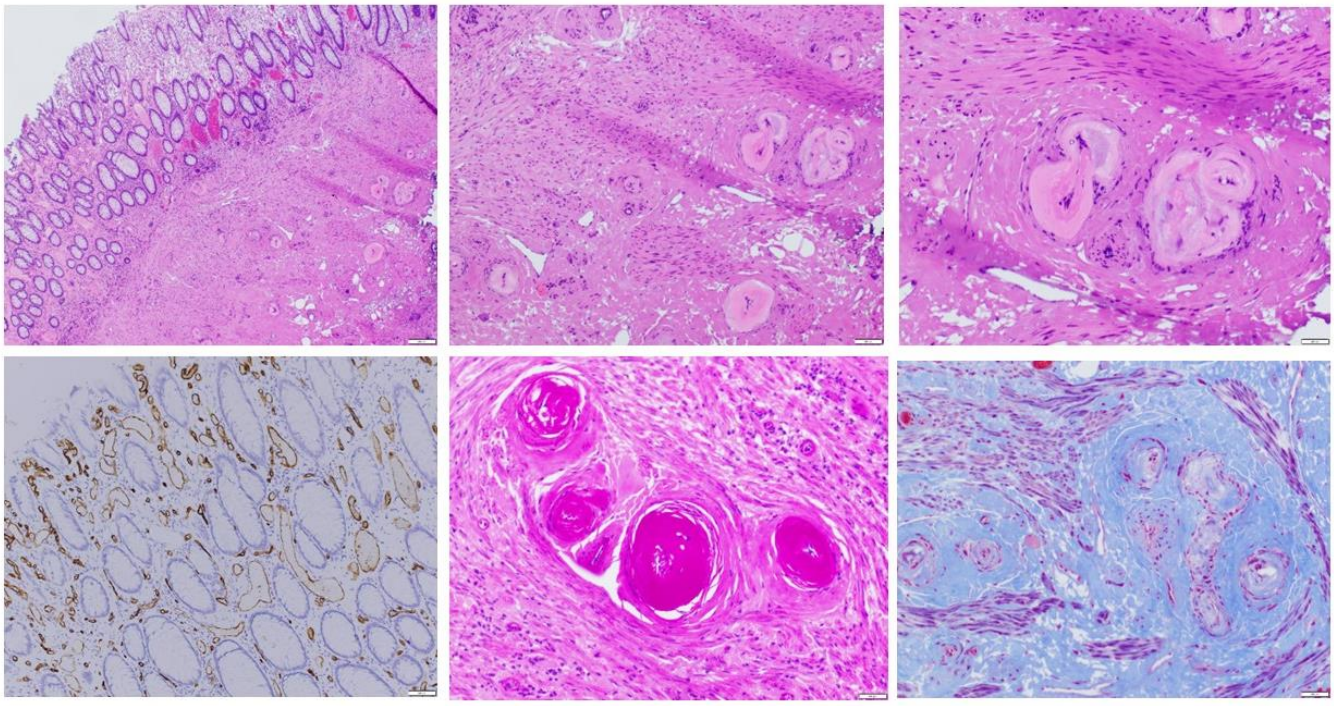

Figure 2. Histology of mucosal and submucosal of small vessels in rectal endoscopic mucosal resection. (**A**) Mucosal ischemic colitis including effacement of superficial epithelium, capillary congestion, hyalinized fibrosis in rectal polyp; (**B**) (10x) and (**C**) (40x) Small artery and vein with intimal and medial hyperplasia and lumen narrowing with H&E stain; (**D**) CD34 immunostain highlighting capillary dilatation and congestion; (**E**). PAS-D stain highlighting the intimal and medial hyperplasia and lumen narrowing; (**F**) Trichrome stain highlighting collagen and fibrosis in small arteries and veins.

Previously, there have only been a few reports of MAVD/V in the bowel presenting as polyp/mass lesions.[2] Later, we reported a case with small bowel necrosis caused with MAVD/V.[4] In that case, small bowel showed extensive ischemic change and small mesenteric arteriovenous wall with smooth muscle and collagen hyperplasia mixed together in intimal and medial layers. From Dr. Patil's report, only three cases were presented as mass/polyps. In our case, the clinician first considered it as a malignant mass (6 cm) with deeper layer invasion. The outside of biopsy pathologist misdiagnosed it as a tubular adenoma due to reparative proliferation after ischemic colitis. In our sample, the large endoscopic mucosal resection makes the diagnosis more dependable. The superficial ischemic and reparative change is mimic tubular adenoma. However, the vascular changes inside of mucosa and submucosa showed an ischemic change and intimal and medial hyperplasia with focal occlusion, which raises a possibility of MSVD/V.

The clinical management approach remains unclear. In our case, the clinical and radiological findings have considered it as a malignant lesion. Luckily, the gastroenterologist did a deeper mucosal resection, which made easier for the diagnosis and avoided further colorectal resection. In future, a multidisciplinary team consisting of vascular surgeons, interventional radiologists, GI pathologists and gastroenterologists should collaborate to develop an individualized treatment plan for the MAVD/V disease. Long-term management and follow-up of patients with MSVD/V should focus on preventing recurrent mesenteric ischemia and preserving bowel function.

In conclusion, we reported a unique presentation of MAVD-like polyp from endoscopic mucosal resection. We name it as submucosal arteriovenous dysplasia.

CONFLICT OF INTEREST DISCLOSURES

The authors have no conflict of interest to disclose.

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