

A Case of Bilateral Acute Inferior Limb Ischemia in a Patient With Ulcerative Colitis

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ABSTRACT: A patient was diagnosed with ulcerative colitis (UC) in 2010. In March 2015, she had abdominal pain, diarrhea, bloody stool, and UC has relapsed. In June 2015, pain and sensory disturbance of both lower limbs appeared. Blood flow at the distal femoral artery was not confirmed with magnetic resonance angiography, and it was diagnosed as bilateral acute inferior limb ischemia. Arterial thrombolectomy with Fogarty's balloon catheter was performed and blood flow was improved. The severity of UC was moderate with Mayo score 8. Thrombosis is considered to be a complication with a high incidence in inflammatory bowel disease. Reports of arterial thrombosis are very rare. It is important to evaluate the risk of bleeding and thrombosis in active or severe cases in UC and need to do thrombotic prophylactic treatment simultaneously with UC treatment.

KEYWORDS: ulcerative colitis, thrombosis

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Introduction

Ulcerative colitis (UC) is a chronic inflammatory bowel disease of an unknown etiology that is localized to the colonic mucosa. Chronic inflammation causes various extraintestinal complications. Venous thrombosis is a relatively common extraintestinal complication, but arterial thrombosis is a rare complication of UC. We report a case of bilateral acute inferior limb ischemia in a patient with UC.

Case Report

A 49-year-old woman was diagnosed as having UC (total colitis and relapsing-remitting types) in 2010. Beginning in March 2015, she had abdominal pain, diarrhea, bloody stool, and UC relapse. She was admitted to another hospital from mid-May 2015 and started induction therapy with prednisolone (PSL) 40 mg/d, salazosulfapyridine 8000 mg/d, and azathioprine 50 mg/d. The symptoms improved, but remission was not attained. In June 2015, pain and sensory disturbance of both lower limbs suddenly appeared. As magnetic resonance angiography (MRA) did not detect the blood flow in the distal femoral artery, she was diagnosed as having bilateral acute inferior limb ischemia. For detailed examination and treatment, she was immediately transferred to our hospital. After hospitalization, anticoagulation therapy was performed with heparin and prostaglandin E1; however, the pain in the lower limb worsened. Thus, arterial thrombolectomy with Fogarty's balloon catheter was performed on the following day. The severe pain was relieved, and the femoral arterial blood flow was improved on magnetic resonance imaging (MRI; Figures 1 and 2). On colonoscopy for detailed evaluation, the severity of UC was moderate, with a Mayo score of 8

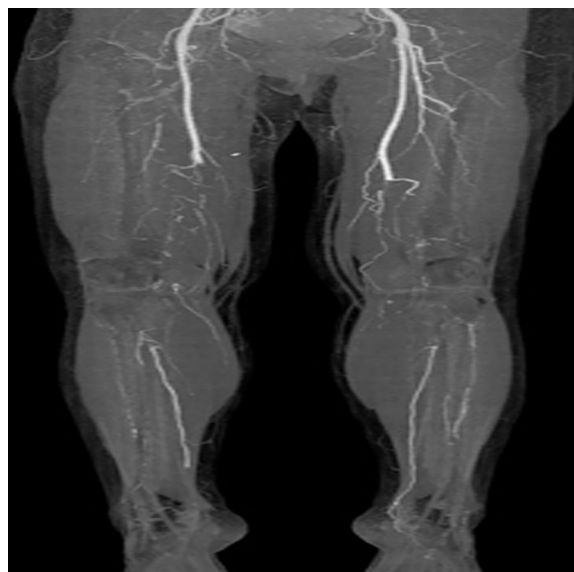


Figure 1. Magnetic resonance angiogram showing no blood flow in the distal femoral artery.

(Figure 3). Therefore, PSL 40 mg/d with salazosulfapyridine 8000 mg/d and azathioprine 50 mg/d were administered. She was discharged on the 28th day from admission, with improvement of arterial thrombosis and UC activity.

Thrombosis is considered a complication with a high incidence of inflammatory bowel disease.¹ Although venous thrombosis is common, arterial thrombosis is a rare condition. The causative factors of thrombosis are thought to be as follows: increased number of thrombocytes and abnormality in the coagulation system according to disease activity. Previous studies showed that an increased number of thrombocytes was



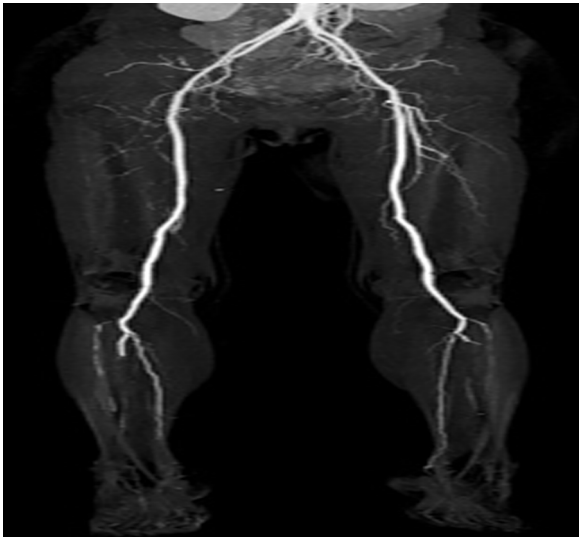


Figure 2. Improvement of distal blood flow from the bilateral femoral artery.

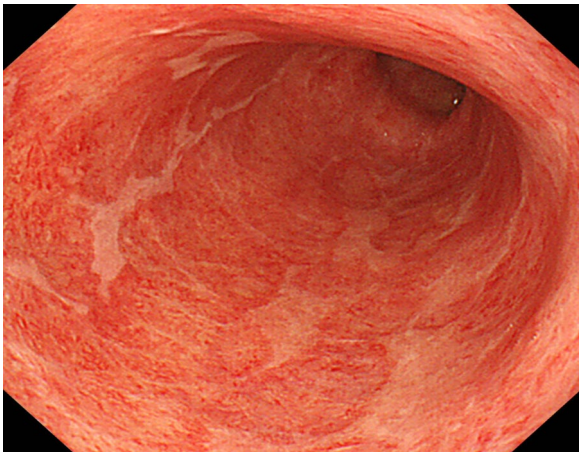


Figure 3. The sigmoid colon with multiple shallow ulcers with a Mayo endoscopic subscore of 3.

more frequently found in active or severe cases of UC.² Of UC cases, 79% that developed thrombosis were in the active phase.³ In the present case, the number of platelets was increased ($64.6 \times 10^4/\mu\text{L}$ on admission) with prolonged inflammation. On the other hand, the coagulation test results were within the normal ranges (prothrombin time/international normalized ratio, 1.10 and D-dimer level, $1.8 \mu\text{g/mL}$). Electrocardiography revealed sinus rhythm, a heart rate of 91 bpm, and no premature atrial/ventricular contraction. Transthoracic echocardiography revealed normal left ventricle function, no left atrium dilatation, and no thrombus. In addition, oral contraception, smoking, hypertension, dyslipidemia, diabetes, autoimmune disease, and malignant tumors as predisposing factors to thrombosis were not identified. Moreover, any accompanying adverse events of PSL administration were unlikely to match the onset time, and no blood coagulation disorder occurred

during treatment of UC. From the examination or patient's background, the main cause of arterial thrombosis in the present case was considered to be the increasing platelet count due to UC activity. The risk of thrombosis tends to increase in active or severe cases of UC, and it is important to evaluate the risk of bleeding and thrombosis and treat thromboprophylaxis at the same time as UC treatment. Also, high levels of factor VIII increase the risk of thrombosis.⁴ It can be said that measuring the activity of fibrinogen and factor VIII and confirming the increase is important for understanding the risk of thromboembolism. In this case, we diagnosed arterial embolism by MRI. An advantage of MRA is that blood flow can be evaluated without using a contrast agent. On the other hand, as for the MRA limitation, when there is a metal such as a pacemaker in the body, imaging may not be possible, imaging may take a long time, and stenosis may be more emphasized due to the influence of resolution. Attention must be paid to thrombotic symptoms during the treatment course and thrombus prophylactic treatments such as mechanical prevention of thrombus by wearing compression stockings or a sequential compression device for the legs, bedside rehabilitation, and administration of anticoagulants after hospitalization.^{5,6} However, the continuous anticoagulant therapy after thromboembolism may increase the risk of bleeding during relapse of UC; thus, along with UC treatment, continuous anticoagulant therapy should be considered if needed.




Author Contributions

MK and TS wrote the manuscript and provided the images. FT, KTa, KTs, KTo, YM, MI, KG, and AI reviewed the manuscript. TS is the article guarantor.

Informed Consent

We had received informed consent from the patient.

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REFERENCES

1. Magro F, Soares JB, Fernandes D. Venous thrombosis and prothrombotic factors in inflammatory bowel disease. *World J Gastroenterol.* 2014;20:4857-4872.
2. Grainge MJ, West J, Card TR. Venous thromboembolism during active disease and remission in inflammatory bowel disease: a cohort study. *Lancet.* 2010;375:657-663.
3. Solem CA, Loftus EV, Tremaine WJ, Sandborn WJ. Venous thromboembolism in inflammatory bowel disease. *Am J Gastroenterol.* 2004;99:97-101.
4. Jenkins PV, Rawley O, Smith OP, O'Donnell JS. Elevated factor VIII levels and risk of venous thrombosis. *Br J Haematol.* 2012;157:653-663.
5. Nguyen GC, Bernstein CN, Bitton A, et al. Consensus statements on the risk, prevention, and treatment of venous thromboembolism in inflammatory bowel disease: Canadian Association of Gastroenterology. *Gastroenterology.* 2014;146:835-848.
6. Lentz SR. Thrombosis in the setting of obesity or inflammatory bowel disease. *Blood.* 2016;128:2388-2394.