

Idiopathic Mesenteric Phlebosclerosis Associated with Long History of Rheumatoid Arthritis: Report of One Case

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1. Abstract

Idiopathic mesenteric phlebosclerosis (IMP) is a rare chronic disease, characterized by thickening of the wall of the right hemi colon with calcification of mesenteric veins. We present a 67-year-old Chinese patient who had a long history of rheumatoid arthritis with taking related drugs for 20 years.

2. Introduction

Idiopathic mesenteric phlebosclerosis (IMP) is a rare entity characterized by chronic intestinal ischemia due to calcification and obstruction of the mesenteric veins. It was first reported by Kayoma et al. in 1991 [1] and proposed “phlebosclerotic colitis” by Yao et al. in 2000 [2], named IMP by Iwashita et al. in 2003 [3]. So far, only more than 200 cases have been reported worldwide. However, the aetiology remains unknown. We experienced such a case, which is typical characteristics of Idiopathic mesenteric phlebosclerosis and was likely associated with the long history of rheumatoid arthritis. After failing conservative treatment, subtotal colectomy was performed. We report this case and also review the relevant literature.

2.1. Presentation of case

A 67-year-old woman was admitted to our gastrointestinal surgical ward with complaints of recurrent abdominal pain and distension for five years. After 2 months of conservative treatment in the local hospital, her symptoms did not ease. She also mentioned difficulty defecation and weight loss (10 kg) within the past year. Physical examination revealed a mass in the lower abdomen. Wrist joints and metacarpophalangeal joints are blocked, fingers and ulna are displaced, swan neck deformities and toes fibula are displaced. All

other body examinations are normal. In a view of signs of thread-like calcifications in the mesenteric veins on computed tomography at local hospital, patient's anamnesis data (she had an oral intake of hydroxychloroquine, methotrexate and methylprednisolone for rheumatoid arthritis for more than 20 years), IMP was suspected.

After admission, further improvement inspections are done. Computed tomography revealed tortuous thread-like calcifications in the ileocolic vein and right colon vein and middle colon vein, ascending colon wall thickening, edema with calcification. (Figure 1). In addition, the left half of the colon collapsed, and diffuse dilation of the small intestine is consistent with mechanical intestinal obstruction. Colon endoscopy revealed purple-blue mucosa extending from the cecum to the transverse colon. Colonoscopy biopsy pathology suggested chronic active inflammation. (Figure 2A) Total gastrointestinal angiography showed a thumb-printing sign, sclerosis of the colonic wall and luminal narrowing of the ascending and the transverse colon (Figure 2B).

Blood tests revealed leukocytosis of $8.9 \times 10^9/L$, CRP 146mg/L, D2 polymers 0.71mg/L. Rheumatoid factor 296.0U/ml (normal, <20U/ml), anticitrullinated protein antibody 106.9U/ml (normal, <20IU/ml), Antinuclear antibody titer 1:100, Anti-O antibody 49.2 U/ml, the erythrocyte sedimentation rate 71mm/hour. Occult blood in stool was positive. Na^+ 113mmol/L, K^+ 2.48mmol/L, Cl^- 75mmol/L, $P_{0.44}$ mmol/L. Her liver function test, renal function test results were normal. The test results combined with the examination findings confirmed the diagnosis of IMP.

On the day 2 after admission, the patient was prescribed TPN assist in correcting internal environment disorders, regulating in-

testinal flora and other treatments. After 1 month symptoms once improved and blood tests normal. However, she presented with recurrence of digestive symptom after enteral nutrition therapy for 1 week. Enteral nutrition therapy failed, so emergency surgery was performed. Surgery exploration showed the wall of the colon was thickening with calcification of mesenteric veins from the cecum to the transverse colon. So we did a subtotal colectomy

and terminal ileostomy. The lesions in the colon was resected and reconstructed. Pathological examination of the resected specimen indicated IMP (Figure 2C).

Two weeks after the operation, the patient was discharged from the hospital. The patient recovered well and was able to eat independently with 5 kg weight gain. After 6 months of follow-up, she was still asymptomatic.



Figure 1: Findings on imaging. A,B,C: Computed tomography revealed showed tortuous thread-like calcifications in the ileocolic vein and right colon vein and middle colon vein, ascending colon wall thickening, edema with calcification.



Figure 2: Findings on imaging: **A:** Colon endoscopy revealed purple-blue mucosa extending from the cecum to the transverse colon. **B:** Total gastrointestinal angiography: a thumb-printing sign, sclerosis of the colonic wall and luminal narrowing of the ascending and the transverse colon. **C:** The wall of the colon was thickening with calcification of mesenteric veins from the cecum to the transverse colon.

3. Discussion

IMP is a rare ischemic colitis. At present, the etiology and pathogenesis of IMP are unclear. The cases are almost concentrated in East Asia, especially Japan, Mainland China and Taiwan [4]. In recent years, an association with herbal medicine has been indicated as one potential cause of this disease, such as geniposide and san-shishi [5,6]. However, our patient did not take herbal medicine for a long time, but for rheumatoid arthritis to take related drugs. After admission, we immediately stopped the drugs. At present, there are no large numbers of reports showing that internal rheumatoid arthritis is related to IMP.

According to reports certain toxins or biochemicals, probably existing in the frequently ingested contents and absorbed to the venous return, may play the most important role in this damage [7]. Our patient has been taking anti-rheumatoid drugs for a long time, which may have caused her illness.

The clinical manifestations and laboratory tests of IMP have no characteristics. A dark purple-colored colon under colonoscopy is a characteristic finding of mesenteric phlebosclerosis [8]. Almost all reported cases shared characteristic radiologic findings in common with multiple fine, tortuous, thread-like or serpentine

calcifications, especially in the right colon [9]. Plain abdominal radiographs showed linear calcifications in the abdomen, especially in the ascending colon [10]. The imaging findings of this patient are consistent with previous reports.

Reviewing the literature, we found that most patients are mild and can be treated conservatively [11]. Our patient has persistent symptoms, and the current conservative treatment has failed. Is surgery treatment needed as the next step? Studies have shown that evaluation of the severity and extent of IMP based on the total mesenteric venous calcification score, number of involved colonic segments, and the presence bowel loop dilatation on CT may be useful to indicate the outcomes of conservative treatment and need for surgery. The article points out that the appropriate cutoff value for number of involved colonic segments was 3.5 and it was 10.5 for the total mesenteric venous calcification score in predicting the need of surgery. But when the total calcification score was 14, it achieved the highest accuracy [12]. According to this scoring method, the total calcification Score of our patients was 14, and the number of colonic segments with mesenteric venous calcification was 3. Therefore, We decided to perform surgical treatment and the surgical method is subtotal colectomy. However, this score

only includes a small number of cases, which needs further study. Cases of laparoscopic subtotal colectomy have also been reported occasionally [13]. Compared with open surgery, no studies have reported to bring better prognosis to patients.

4. Conclusion

As described, IMP is very rare and has no specific symptoms. The diagnosis is confirmed based on imaging examinations suggesting characteristic calcification of mesenteric line and histology suggesting mesenteric vein wall calcification. The treatment is mostly conservative. If the symptoms are obvious and conservative treatment is ineffective, surgical treatment should be performed. It is very important of appropriate preoperative assessment for accurately assessing the IMP range and determining the optimal excision range.

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