

Multiple chronic non-specific ulcer of small intestine characterized by anemia and hypoalbuminemia

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Abstract

A female patient with anemia and hypoalbuminemia was admitted to our hospital due to an over 20-year history of recurrent dizziness, fatigue and ankle edema. She was diagnosed as multiple chronic non-specific ulcer of the small intestine characterized by non-specific histology and persistent gastrointestinal bleeding.

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INTRODUCTION

With the wide use of capsule endoscopy, small intestinal ulcer associated with chronic bleeding is commonly seen in clinical practice. However, its diagnosis and treatment are complicated. After the causes of ulcer such as Crohn's disease, Behcet's disease and tuberculosis are excluded, most cases are diagnosed as "idiopathic chronic ulcerative enteritis"^[1]. Since 1960, cases of non-specific ulcer of small intestine have been reported^[2-4]. Most of them were caused by non-steroidal anti-inflammatory drugs (NSAID) or potassium tablets. Matsumoto *et al*^[5,6] have reported a multiple chronic non-specific ulcer of small intestine (CNSU), which is not related to NSAID. We, here, report a case of multiple chronic non-specific ulcer of the small intestine characterized by non-specific histology and persistent gastrointestinal (GI) bleeding.

CASE REPORT

A 42-year-old female was admitted to our hospital due to an over 20-year history of recurrent dizziness, fatigue and ankle edema. Over the past 20 years, she had dizziness, fatigue and abdominal distension without any identifiable causes. At the same time, she developed ankle edema which resolved spontaneously after rest. Laboratory test at a local hospital showed that she had severe iron deficiency anemia [hemoglobin (HB) 27 g/L] and hypoalbuminemia (albumin 11 g/L). Her symptoms could always be relieved temporarily after intravenous infusion of albumin and packed red blood cells. However, over the past 4 mo, she had repeated episodes of chest tightness and palpitation after activities. She also complained of intermittent abdominal pain after meal but had no fever, nausea or vomit. Her bowel movements were normal except for occasionally mild diarrhea. She denied any decreased appetite, nausea and vomiting, hematemesis, hematochezia, melena, tenesmus and stools with mucus or pus.

She had no known history of hepatitis or chronic renal disease or smoking or alcohol, and exposure to contaminated water. She denied any use of NSAID. Her parents were healthy and her little brother was dead at the age of 10 years due to unknown cause. Her son and husband were healthy.

Physical examination demonstrated that her temperature was 36.8°C, blood pressure was 92/60 mmHg, respiration was 18/min, and heart rate was 92 bpm. The patient was alert and fully oriented but appeared pale, cold and clammy. She had no jaundice, liver palm or spider angioma, eruption or purpura on the skin. No superficial lymph nodes could be palpated. Her trachea was slightly shifted to the left with no jugular venous distention. Her right chest wall movement and tactile fremitus over the right lung base were decreased with absent breath sounds and dullness to percussion. Her heart had a grade 2/6 diastolic murmur at the apex. An abdominal bulge and positive shifting dullness were found with active bowel sound. No abdominal wall vein dilatation, tenderness or rebound tenderness were found. No mass, liver and spleen were palpable. Digital rectal examination was negative. She had moderate pitting edema of the lower extremities.

Her white blood cells were $8.0 \times 10^{12}/L$, HB was 59 g/L, mean cell volume was 65 fL, mean corpuscular hemoglobin was 18.1 pg, and platelets were $445 \times 10^9/L$. Her TB/CB was 0.15/0.07 mg/mL, A/G was 1.3/2.0, alanine aminotransferase was 25 U/L, aspartate aminotransferase was 20 U/L, reticulocytes was 2.9%, erythrocyte sedimentation rate was 28 mm/h. Bone marrow aspiration showed proliferation of erythrocyte lineage and mature red blood cells with an increased central pallor. Iron staining revealed 2% intracellular iron but no extracellular iron. No proteinuria or haematuria was detected at a urinalysis. Renal function was normal. Rheumatoid factors and full antinuclear autoantibodies were negative. Serological tests were negative for hepatitis B virus, hepatitis C virus and human immunodeficiency virus. Her serum K^+ and Na^+ were 3.15 mEq/L and 133 mEq/L, respectively. Abdominal ultrasonography showed the presence of moderate ascites and a right pleural effusion. Fecal occult blood test (FOBT) was positive while stool culture was negative. Both upper GI endoscopy and colonoscopy were negative. Capsule endoscopy showed multiple, sharply demarcated ulcers limited in the ileum with a circular shape. The margins of ulcers were clear and the intervening mucosa was normal. Stenosis could also be seen in the ileum.

The patient was treated with intravenous infusion of albumin and packed red blood cells as well as intramuscular injection of iron. After 4 wk of treatment, the dizziness and abdominal distension were gradually improved and lower extremity edema also receded. Hb increased temporarily to 90 g/L and abdominal ultrasound showed only a small amount of ascites and pleural effusion. However, FOBT remained positive and abdominal pain did not relieve. Oral prednisone was given for 1 wk but did not relieve the pain. A laparotomy was proposed

but she refused. She was discharged and scheduled for outpatient follow-up.

DISCUSSION

GI bleeding-induced anemia is the most typical presenting symptom of patients suffering from CNSU and low serum protein concentration is also seen^[5]. Our patient had pronounced anemia and hypoalbuminemia. Her initial manifestation was pronounced anemia followed by ascites and pleural effusions. The diagnostic criteria for CNSU were established as previously described^[6], including persistent anemia for more than 1 year, small intestinal ulcers, absence of clinical evidence suggestive of mycobacterial infection, absence of clinical evidence suggestive of Crohn's disease, and lack of any dermatologic, ophthalmologic or genital symptom suggestive of Behcet's disease. The ulcers in our patient were different from those of Crohn's disease characterized by longitudinal ulcers and cobblestone appearance. There was also no evidence of complication suggestive of Crohn's disease because no perforation and fistulisation were found although the patient had a very long course of disease. Behcet's disease could also be excluded since there was no clinical evidence showing dermatologic, ophthalmologic or genital symptom in our patient.

Capsule endoscopy confirmed the diagnosis of CNSU in our patient. Matsumoto *et al.*^[5] reported that ulcers in CNSU patients are predominantly found in the ileum, which are circular or irregular in shape. The margins of ulcers are always clear and the intervening mucosa appears normal. Capsule endoscopy showed typical circular ulcers limited in the ileum of our patient, which is consistent with the reported findings^[5].

The clinical and endoscopic features of CNSU are similar to those of NSAID-induced enteropathy. Matsumoto *et al.*^[6] compared the enteroscopic findings in CNSU and NSAID-induced enteropathy, and found that both are characterized by concentric stenosis and ulcers with non-specific histology while the lesions of small intestine are different in respect to their site and stage. CNSU patients have active, sharply demarcated ulcers limited in ileum while few ulcers are found in NSAID-induced enteropathy. Since our patient had no history of NSAID use, the possibility of NSAID-induced enteropathy could be excluded.

CNSU is different from another idiopathic small intestinal multiple ulcer disease described as cryptogenic multifocal ulcerous stenosing enteritis (CMUSE)^[7], which is an independent, rare and poorly understood disease characterized by non-specific small intestinal ulceration and stenosis which responds to corticosteroid therapy^[7]. Perlemuter *et al.*^[7] described that CMUSE syndrome is characterized by chronic diarrhea, bouts of intestinal obstruction, and ulcerative stenosis of the small intestine. A very important feature of CMUSE is that patients respond dramatically to corticosteroid therapy^[7]. However, the therapeutic effect of corticosteroid in our patient was not good. Another dif-

ference is that anemia and hypoalbuminemia are not often seen in CMUSE patients. Our patient had a long history of pronounced anemia and hypoalbuminemia prior to the development of abdominal pain, suggesting that stenosis may not develop rapidly in CNSU.

Capsule endoscopy is the best diagnostic tool for obscure GI bleeding. Our patient was admitted because of her chronic GI bleeding with unknown origin. Since upper GI endoscopy and colonoscopy showed negative results, capsule endoscopy showed multiple circular ulcers. The affected site was limited in the ileum, thus providing the most important evidence for the diagnosis of our patient.

In conclusion, the pathophysiology of CNSU remains poorly understood. CNSU is a disease responsible for obscure GI bleeding arising from the small intestine. Capsule endoscopy contributes to the diagnosis of this peculiar form of small intestine disease.

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REFERENCES

- 1 **Robertson DA**, Dixon MF, Scott BB, Simpson FG, Losowsky MS. Small intestinal ulceration: diagnostic difficulties in relation to coeliac disease. *Gut* 1983; **24**: 565-574
- 2 **Graham DY**, Bynum TE. Primary nonspecific small bowel ulceration as a source of chronic bleeding. Report of a case and review of the approach to localization of the site of small bowel hemorrhage. *Am J Gastroenterol* 1974; **62**: 350-355
- 3 **Glynn MJ**, Pendower J, Shousha S, Parkins RA. Recurrent bleeding from idiopathic ulceration of small bowel. *Br Med J (Clin Res Ed)* 1984; **288**: 975-976
- 4 **Wilson IR**, Cooley NV, Luibel FJ. Nonspecific stenosing small bowel ulcers. Experience in one California county. *Am J Gastroenterol* 1968; **50**: 449-455
- 5 **Matsumoto T**, Iida M, Matsui T, Yao T, Watanabe H, Yao T, Okabe H. Non-specific multiple ulcers of the small intestine unrelated to non-steroidal anti-inflammatory drugs. *J Clin Pathol* 2004; **57**: 1145-1150
- 6 **Matsumoto T**, Nakamura S, Esaki M, Yada S, Koga H, Yao T, Iida M. Endoscopic features of chronic nonspecific multiple ulcers of the small intestine: comparison with nonsteroidal anti-inflammatory drug-induced enteropathy. *Dig Dis Sci* 2006; **51**: 1357-1363
- 7 **Perlemuter G**, Guillevin L, Legman P, Weiss L, Couturier D, Chaussade S. Cryptogenetic multifocal ulcerous stenosing enteritis: an atypical type of vasculitis or a disease mimicking vasculitis. *Gut* 2001; **48**: 333-338

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