Cryptogenic multifocal ulcerous stenosing enteritis (CMUSE) is a rare chronic and recurrent disease with unknown etiology. It is characterized by shallow ulcers, submucosa thickness, and no signs of systemic inflammation. Now, steroids are the principle therapy for CMUSE. However, still, there were few patients who did not respond to it. Here, we present a young woman who diagnosed with CMUSE. She underwent a surgery because of capsule endoscopy retention. After relapse, she had received budesonide and methylprednisolone separately while both of them did not stop her disease. Till now, 24 years after her initial symptom, there are no signs of carcinoma yet.

Keywords: Cryptogenic multifocal ulcerous stenosing enteritis, intestinal ulceration, steroid-resistant

Case Report

A Steroid-resistant Cryptogenic Multifocal Ulcerous Stenosing Enteritis

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Fecal occult blood test was positive. A laboratory test confirmed hypoalbuminemia (20.8 g/L), iron deficiency anemia (hemoglobin at 66 g/L), and slightly increased C-reactive protein level (8.43mg/L). Her renal and liver functions, erythrocyte sedimentation rate, immunoglobulin level, complement level, and autoimmune antibodies were within normal range. (PPD) purified protein derivative skin test was negative. Gastroscopy showed mild gastritis, and colonoscopy was normal. Abdominal computed tomography revealed thickened small intestinal wall. On capsule endoscopy, multiple shallow ulcers and stenosis were found in the small intestine. Exploratory laparotomy was performed for capsule retention. In operation, several strictures located between 130 and 50 cm proximal to ileocecal valve were found and the minimal distance between two strictures was about 3 cm. The stricture lesions were removed. Postoperative pathology showed nonspecific chronic inflammatory, superficial ulcers restricted in the mucosa, and submucosa fibrosis. No granulomas, lymphadenopathy, or vasculitis was found [Figure 1]. She received symptomatic treatment and discharged without further therapy.

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From then on, she was readmitted to our hospital three times for the same reason and along with low albumin, anemia. Following enteroscopy found circular and irregular superficial ulcers with multiple stenosis 60 cm to ileocecal valve [Figure 2]. Biopsy still showed nonspecific moderate chronic inflammation and focal erosion. Acid-fast stain and polymerase chain reaction for tuberculosis were negative. CMUSE was diagnosed. We first prescribed budesonide (9 mg/day) for 1 month; she had lasted abdominal pain and melena. Then, methylprednisolone (40 mg/day) was tried and did not improve her symptom. Steroids were discontinued. She refused further therapy, except symptomatic treatment. Now 1 year after her last hospitalization, she still had continued tarry stool.

**CONCLUSION**

We reported a steroid-resistant CMUSE patient with continued tarry stool for almost 24 years. More studies are needed to find the cause of CMUSE and the therapy for steroid-resistant ones. Fortunately, no CMUSE-related cancer or death has been reported. In this situation, symptomatic treatment may be a choice for noninvasive treatment.

**Declaration of patient consent**

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

**Financial support and sponsorship**

Nil.

**Conflicts of interest**

There are no conflicts of interest.

**REFERENCES**


