A rare case of simple ulcer of the colon in a 7-year-old girl is reported. Simple ulcer is clinically and pathologically recognized as a serious disease linked to intestinal Behcet’s disease. Recently, some immunomodulators, such as thalidomide and antitumor necrosis factor monoclonal antibody, have been used to treat Behcet’s disease, with varying degrees of success. Pentoxifylline (PTX) is also known to inhibit such inflammatory mediators as tumor necrosis factor-α, interleukin-1β, and interleukin-6. In this present patient combination treatment with prednisolone, azathioprine, and PTX improved corticosteroid dependence palliatively and prevented further relapse during a follow-up period of more than 12 months, without serious side effects.

Key words: simple ulcer, intestinal Behcet’s disease, pentoxifylline, cytokine, child

Introduction

Simple ulcer of the colon is uncommon, especially in childhood, and its etiology has yet to be elucidated.1 Owing to recent advances in colonoscopic techniques, this lesion is found frequently in daily clinical practice.1–5 However, differentiating this disease from Behcet’s ulcer is often impossible pathologically, and the clinical course of simple ulcer may also prove difficult to manage because of resistance to conservative internal treatments and a high recurrence rate after surgical resection.1–3,6 This article describes a 7-year-old girl with simple ulcer of the colon, who was effectively treated with prednisolone, azathioprine, and pentoxifylline (PTX), which is known as an inflammatory cytokine inhibitor.

Report of a case

A seven-year-old girl was admitted to our hospital in May 1993 complaining of prolonged abdominal pain, of 11 days’ duration, along with fever and recurrent vomiting. The pain gradually became sharper, with abdominal cramps. Her past medical history included acute enterocolitis at the age of 2 years and necrotic gingivitis at the age of 5 years. She had completely recovered from both diseases with conservative treatment. At the initial examination, she weighed 24.3 kg, her body temperature was 38°C, and abdominal tenderness was found in the right lower quadrant, without guarding, while moderate dehydration was also observed. A painful and recurrent oral aphthous ulcer was observed across her second right molar tooth. However, no ulcerating genital or anal lesions, skin eruptions, or inflammatory ocular lesions were seen.

Laboratory studies revealed the white blood cell count to be 18 300/mm³, with 84% neutrophils and 8% lymphocytes, and hematocrit and hemoglobin were slightly elevated due to dehydration. Although her serological findings demonstrated a C-reactive protein level of 14.3 mg/dl and a mildly elevated erythrocyte sediment rate, chemical analysis and urinalysis findings were all within the normal limits. Stool was positive for occult blood, and no pathologic bacteria were detected by a stool culture. HLA-B51 antigen, needle, and tuberculin reaction findings were all negative.

When massive anal bleeding with abdominal cramps occurred on the fifth day, colonoscopic examination up to the splenic flexure revealed multiple ulcer scars and pseudopolyps in the rectum, sigmoid colon, and descending colon. Crohn’s disease was suspected, and she was treated with prednisolone (1 mg/kg per
day by month) and salazosulfapyridine (1.5 g/day). After the treatment, her abdominal pain and fever disappeared and the C-reactive protein concentration immediately decreased, to less than 0.5 mg/dl in 2 weeks.

However, after the decreasing of prednisolone, a relapse of abdominal pain and anal bleeding occurred, in July 1993. Colonoscopy was therefore performed again and revealed round and oval ulcers, measuring from 2 to 3 mm to several centimeters in size, all over the colon. The main lesion in the ileocecal area was a large, bleeding punched-out ulcer (Fig. 1). Histologically, there was a nonspecific inflammatory reaction with infiltrating neutrophils and lymphocytes (Fig. 2). No granulomas were noted. According to the endoscopic and histologic findings, the ulcerative lesions were clinically diagnosed as simple ulcer of the colon.

From the second admission in July 1993, prednisolone (1.5 mg/kg per day) was restarted, and high-dose immunoglobulin therapy (400 mg/kg per day, for 3 consecutive days, i.v.), with total parenteral nutrition for 5 weeks, followed by enteral nutrition, was performed. After receiving azathioprine (100 mg/day, by month), the patient was changed from enteral nutrition to a

Fig. 1a,b. Colonoscopic findings 5 months after the onset. a The colonoscopic picture in the ileocecal area demonstrates an active ulcer, several centimeters in diameter. b The picture at a proximal distance shows that a thick, white slough with bleeding covers the ulcer.

Fig. 2a,b. Histological findings show nonspecific inflammatory reaction with infiltrating neutrophils and lymphocytes. No granulomas were noted.