INTRODUCTION

Though gastrointestinal symptoms are relatively common in BD, ulcerative change of the intestine is not that frequent. The most common sites for intestinal BD are the terminal ileum and the cecum. There are reports of fistula formation in BD. In those, some are related to surgery and others are not. BD with cecocoeal fistula, possibly associated with a past operation, has not been reported in the literature.

CASE

A 38-year-old man presented with massive, bloody diarrhea and colicky pain in RLA. He had been having recurrent oral ulcer, genital ulcer and erythema nodosum-like lesion for the past ten years. Six years ago, he had an appendectomy done at a primary clinic. At that time, multiple ulcers were noted in the ileocecal region and histology showed minimal inflammation in the appendix. He was referred to the university medical center and diagnosed as having an intestinal BD. He had been treated with low-dose prednisolone and sulfasalazine. After surgery, intermittent, severe abdominal pain occurred while he had been on medication on and off.

On examination, he had a regular pulse of 120 beats/min, blood pressure of 70/40 mm Hg and body temperature at 36.5°C. His conjunctiva was mildly pale. The lungs were clear to auscultation. Though his abdomen was soft, the abdominal examination revealed tenderness and rebound tenderness in RLA. He had a hematocrit of 33.5%, a white blood cell count of 11.8 $10^9$/mm$^3$ and a platelet count of 185 $10^9$/mm$^3$. The antinuclear antibody, rheumatoid factor and antineutrophil cytoplasmic antibody were negative. Plain abdomen showed no specific findings except for the focal ileus in RLA.

Even with enough saline and transfusion of packed red blood cells, he had persisting hypotension and severe abdominal pain. Emergency ileocecal segmental resection was performed. Pathologic examination revealed large cavitating ulcer (figure 1), lymphocyte aggregates and lymphocytic vasculitis (figure 2) in the ileocecal region. There was cecocoeal fistula (figures 3 & 4), presumably around the previous appendectomy site.

He was started on oral prednisolone 10 mg/day, sulfasalazine 2 g/day and cyclophosphamide 100 mg/day.

Key Words: Behcet’s disease, Cecocoeal fistula, Ileocecal ulcer, Appendectomy
in the sixth postoperative day. His postoperative course was uneventful.

DISCUSSION

BD is a multisystemic disorder characterized by oral ulcer, genital ulcer, uveitis and skin lesions, most likely occurring with the underlying vasculitis. There is other organ involvement of joints, heart and lungs, as well as neurologic and gastrointestinal involvement. The etiology remains unclear. Genetic and environmental factors probably have a role in the pathogenesis. There are no specific diagnostic or laboratory tests for BD. Diagnosis depends upon the proper history and clinical manifestations. Our case fulfilled the diagnostic criteria of the Intestinal Study Group for Behcet's disease.

Many patients complained of gastrointestinal symptoms such as nausea, vomiting and abdominal pain, but the ulcerative changes in the intestine were found in 1% or less of all patients with BD. The commonest sites for the ulcerative changes of intestinal BD were terminal ileum in 44%, followed by the ileocecal region in 34% and the...
The cecum in 12%,. The clinical manifestations of intestinal BD are similar to inflammatory bowel disease, especially Crohn’s disease. But lymphoid aggregates, submucosal fibrosis, no granuloma, and deep penetrating, easily perforating ulcers in Behcet’s colitis help to distinguish it from Crohn’s disease. Histology in our case revealed lymphoid aggregates, no granuloma and deep penetrating ulcers in the ileocecal region. About 22% of the patients with intestinal BD developed symptoms mimicking appendicitis during the clinical course. Because the most common sites of intestinal BD are the terminal ileum and the ileocecal region, some cases can be diagnosed as an appendicitis. Our case had a past history of erroneous diagnosis with an appendectomy. The opening of the cecocoeal fistula was around the site of the appendectomy. The exact mechanism of fistula formation in BD is unclear. We could speculate that vasculitis and ulcer necrosis, along with pathergy reaction by surgical trauma in our case, might have contributed to the fistula formation.

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