Abstract We report a 27-year-old female with Crohn’s disease clinically misdiagnosed with intestinal endometriosis. Her complaints were abdominal pain and fullness, which occurred monthly during her menstrual period. Although we had no histopathological evidence, we diagnosed her as bowel endometriosis on the basis of her clinical course. Since nafarelin acetate therapy started, the symptoms due to mechanical subileus have improved. The transverse colon, a 70 cm segment of the ileum, including the terminal ileum, were resected because of repeated symptoms of bowel obstruction despite prolonged nafarelin therapy. Histopathological findings of the resected specimen revealed Crohn’s disease without endometrial tissue. In our patient, an increased cortisol and ACTH secretion, a side effect of nafarelin, was noted during the therapy. This case showed that nafarelin therapy could increase serum concentration of ACTH and cortisol, which may have incidentally exerted anti-inflammatory effects on Crohn’s disease. This case showed that some female patients with Crohn’s disease might be misdiagnosed as having intestinal endometriosis because of a reduction in symptoms by nafarelin therapy. We describe a relationship between nafarelin acetate and Crohn’s disease in this case report with a discussion about possible mechanisms for this drug’s action.

Key words Crohn’s disease · Bowel endometriosis · Nafarelin acetate

Introduction

Endometriosis may involve abdominal and pelvic organs, including ovaries, fallopian tubes, and rarely the bowel. Invasive bowel endometriosis can present as a bowel obstruction in an acute, chronic, or intermittent nature [4, 9]. The major cause of obstruction is stricture formation and adhesions, which occasionally mimic Crohn’s disease or a neoplasm in the clinical presentation [1, 2, 5, 7]. For the treatment of bowel endometriosis, nafarelin acetate, a gonadotropin-releasing hormone (GnRH) analogue, is often used. We report a case of Crohn’s disease clinically diagnosed as intestinal endometriosis, due to a reduction of symptoms following treatment with nafarelin acetate. In our patient, increased cortisol secretion, a side effect of nafarelin, may have incidentally exerted anti-inflammatory effects on Crohn’s disease. This case showed that some female patients with Crohn’s disease might be misdiagnosed as having intestinal endometriosis because of a reduction in symptoms by nafarelin therapy. We describe a relationship between nafarelin acetate and Crohn’s disease in this case report with a discussion about possible mechanisms for this drug’s action.

Case report

In June 1998, a 27-year-old Japanese woman was admitted to our hospital complaining of perimenstrual lower abdominal pain and fullness, which occurred monthly during her menstrual period since 1994, and gradually increased in severity. She had no history of a sexually transmitted disease or surgery. No relatives had an inflammatory bowel disease or endometriosis. She had delivered 2 healthy babies before bilateral salpinges ligations were performed at 21 years of age.

A physical examination on admission revealed a thin and pale woman weighing 50 kg. Her blood pressure was 110/60 mmHg. The abdomen was soft and distended with periumbilical tenderness and fullness. There was neither hepatosplenomegaly nor lymphoadenopathy. She demonstrated no arthralgia. Skin rash and erythema nodosum were not found. There was no perianal fistula. A pelvic examination revealed no cervical motion tenderness and fullness. There was neither hepatosplenomegaly nor lymphoadenopathy. She demonstrated no arthralgia. Skin rash and erythema nodosum were not found. There was no perianal fistula. A pelvic examination revealed no cervical motion tenderness, adnexal tenderness, or masses. Results of laboratory studies on admission showed: white blood cell count, 4800/µL; red blood cell count, 308x10^6/µL; hemoglobin, 8.6 g/dL; C-reactive protein 2.6 mg/dL; albumin, 3.0 mg/dL; carcinoembryonic antigen, 0.8 ng/mL; and CA125, 14 U/mL.

A nafarelin acetate test showed an extrinsic nodular filling defect in the terminal ileum, a...
5 cm narrow segment in the sigmoid colon, and a poorly mobile annular stricture of the transverse colon. Neither longitudinal ulcer nor inflammatory polyps were noted. Colonoscopy demonstrated a cobblestone appearance in the sigmoid colon and an ulceration in the transverse colon. Histopathological findings of the biopsied specimen disclosed chronic mucosal and submucosal inflammation with no granulomas or endometrial tissue.

Despite no pathological evidence, her home doctor clinically diagnosed her with bowel endometriosis based on interviews and the clinical course. Since nafarelin acetate therapy was started, the symptoms due to mechanical ileus have improved. Although we also had no pathological evidence, the good response to nafarelin therapy encouraged us to make such a diagnosis and treatment. She had 9 similar and more severe episodes of perimenstrual subileus over a 3-year period, which improved under nafarelin therapy, whereas subileus often occurred during the rest of the therapy. After obtaining adequate informed consent, laparotomy was performed due to repeated symptoms of small bowel obstruction despite prolonged nafarelin therapy.

At laparotomy, extensive adhesion and fistula formation between the terminal ileum and the transverse colon were found. The terminal ileum was inflamed, indurated, markedly thickened, and angulated. The uterus, Fallopian tubes and ovaries were normal. There was no stricture or induration within other segments of the alimentary tract. The transverse colon and a 70 cm segment of the ileum, including the terminal ileum, were resected. An ileocolic anastomosis was performed.

**Gross findings.** Macroscopically, a longitudinal ulceration was observed in the anal side of the stenotic lesion of the terminal ileum. A fistula was detected between the transverse colon and the ulcer scar in the terminal ileum. A cobblestone appearance was noted in the mucosa of the transverse colon.

**Microscopic findings (Fig. 1).** Microscopic examination of the resected terminal ileum revealed marked ulcers and fissures with transmural inflammatory cell infiltration and fibrosis. Non-caseous granulomas were found in both the terminal ileum and the transverse colon, compatible with Crohn’s disease. No endometrial tissue was noted in any segment of the resected specimen.

After obtaining a histopathological diagnosis, we measured plasma concentrations of adrenocorticotropic hormone (ACTH) and cortisol using frozen and stored plasma taken in the morning. ACTH and cortisol during nafarelin therapy were 34.6 pg/mL.