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Ileal J-Pouch Perforation: Case Report

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Rezumat

Anastomoză ileo-anală cu rezervor ileal în J perforat - prezentare de caz

Un pacient în vârstă de 34 ani, supus în urmă cu 5 ani unei operații de colectomie totală cu anastomoză ileo-anală cu rezervor ileal în J, ca urmare a unui diagnostic de polipoză adenomatoasă familială, a fost internat în regim de urgență pentru acuze de durere abdominală intensă datând de aproximativ 4 zile. Examenul fizic a revelat durere difuză la palparea abdominală, în mod special în cadranele inferioare. Ecografia abdominală a decelat prezența de lichid între ansele intestinale, iar examenul CT a decelat niveluri hidroaerice libere în abdomen. În timpul laparotomiei efectuate pentru lărgirea anastomozei ileo-anale la aproximativ 12 cm diametru, o perforație de 2 mm a fost decelată la nivelul capătului orb al anastomozei. A fost rezolvată prioritar perforația, după care s-a efectuat ileostomă de protecție. În timpul endoscopiei postoperatorii nu s-au observat obstrucție sau stază, dar a fost decelată inflamație la nivelul rezervorului ileal în J. Pacientul a fost externat la 20 de zile de la intervenția chirurgicala și urmărit postoperator endoscopic. Rezultatele endoscopiei efectuate la 6 luni de la operație au fost normale.

Cuvinte cheie: polipoză adenomatoasă familială, anastomoză ileo-anală, perforație intestinală, rezervor ileal în J, inflamația rezervorului ileal

Abstract

A 34-year-old male patient who had undergone total colectomy and J-pouch ileanal anastomosis subsequent to diagnosis of familial adenomatous polyposis five years previously was admitted to the emergency room with complaints of severe abdominal pain of a four-day duration. Physical examination revealed widespread tenderness throughout the abdomen, especially in the lower quadrant. Abdominal ultrasonography revealed fluid between intestinal loops and computed tomography revealed free air and fluid in the abdomen. During laparotomy to expand the ileal J-pouch to approximately 12 cm in diameter, a 2-mm perforation was detected in the blind end of the ileal J-pouch. The perforation was repaired primarily and protective ileostomy was performed. During postoperative endoscopy, neither obstruction nor stasis was observed, but pouchitis was observed in the ileal J-pouch. The patient was postoperatively discharged on the 20th day and followed endoscopically. The endoscopic findings were normal in the sixth month postsurgery.

Key words: familial adenomatous polyposis, ileoanal pouch, ıntestinal perforation, J-pouch, pouchitis

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Introduction

Familial adenomatous polyposis (FAP) is a rare autosomal

dominant syndrome constituting 1% of cases of colorectal adenocarcinoma and mainly affecting individuals with a known family history. The clinical presentation of the disease is development of hundreds or thousands of adenomatous polyps immediately after puberty. In FAP patients, the lifetime risk of colorectal cancer reaches 100% at 50 years of age, and almost 100% develop cancer unless prophylactic colectomy is performed. FAP may be associated with certain nonintestinal symptoms, including congenital hypertrophy of the retinal pigment epithelium, desmoid tumors, epidermoid cysts, osteomas of the mandible (Gardner Syndrome), and central nervous system tumors (Turcot syndrome) (1). In the surgical treatment of FAP patients, total proctocolectomy and ileal pouch–anal anastomosis have become the most widely practiced options. Among the treatment options, J-pouch ileoanal anastomosis is preferred because it can be performed easier and has better functional results compared to other options (2). In 3–15% of patients with an ileal pouch, functional capacity of the pouch is lost due to various complications (3). This case report describes the presentation and treatment of J-pouch ileal perforation, a rarely observed complication, in a patient with perforation of ileal J-pouch blunt ends.

Case report

A 34-year-old male patient who had undergone total colectomy and J-pouch ileanal anastomosis subsequent to diagnosis with familial adenomatous polyposis five years previously was admitted to the emergency room with complaints of continuous severe abdominal pain of a four-day duration. The patient complained of frequent defecation (6–8 times/day) that often took the form of diarrhea. Physical examination revealed widespread tenderness throughout the abdomen and especially in the lower quadrant. Although outpatient direct abdominal radiography was normal, abdominal ultrasonography revealed intense content liquid and free air echoes between dilated bowel loops. Computed tomography revealed free air and fluid in the abdomen. Laboratory testing revealed a white blood cell count of 14800/mm^3, a C-reactive protein level of 145 mg/dl, and liver and kidney parameters within normal values.

Subsequent performance of laparotomy revealed intestinal content and fluid in the abdominal cavity. Examination of intra-abdominal bowel segments revealed purulent fluid, abscess formation, and fibrous adhesions, providing evidence of severe peritonitis. Widening of the ileal J-pouch to approximately 12 cm in diameter revealed approximately 2 mm of perforation in the blind end of the ileal J-pouch (Fig. 1). The perforation was repaired primarily and protective right proximal loop ileostomy was performed. Postoperative endoscopy revealed no evidence of obstruction or stasis. While no polyps were observed in the ileal J-pouch, histopathological examination revealed findings consistent with pouchitis in the ileal J-pouch. Postsurgery metronidazole (15–20 mg/kg/day) and ciprofloxacin (1000 mg/day) treatment was initiated and continued for weeks. Systemic examination of the patient revealed a soft tissue tumor on the right mandible of the face. Ophthalmologic examination revealed hypertrophy of the bilateral congenital retinal pigment epithelium in the fundus. Color fundus images revealed gray granular pigmentation in the lesion and areas of depigmentation (Fig. 2). Based on these findings, Gardner's syndrome was suspected. The patient was postoperatively discharged on the 20th day of hospitalization and continued undergoing endoscopic follow-up. Endoscopic examination in the second month postsurgery revealed a decrease in the severity of the pouchitis findings accompanied by improvement in the clinical symptoms of pouchitis. After obtaining normal postoperative examination findings in the sixth month postsurgery, ileostomy closure was planned.

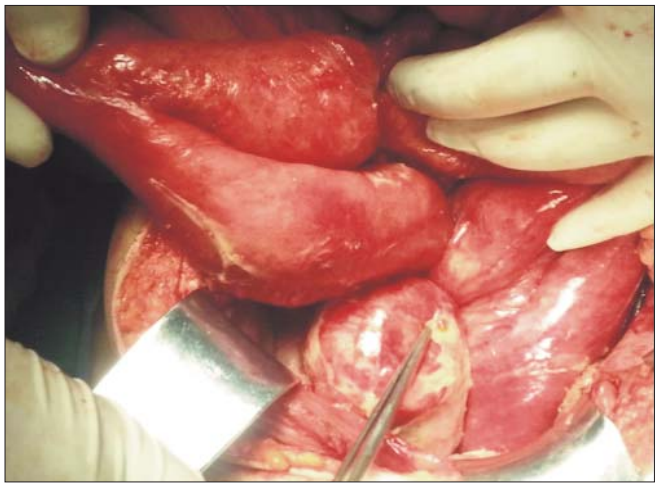

Figure 1. *Ileal j-pouch perforation*

Discussion

J-pouch ileoanal anastomosis is a safe surgical technique with minimal incidence of perioperative mortality and morbidity widely that is performed on patients with a diagnosis of

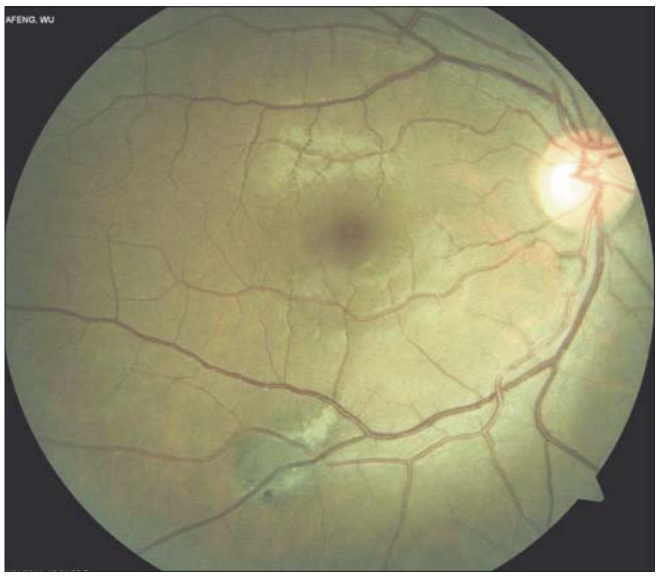

Figure 2. *Congenital hypertrophy of the retinal pigment epithelium*

familial polyposis coli or ulcerative colitis. However, several mechanical, inflammatory, functional, and metabolic complications have been associated with ileal pouch surgery. The most common causes of loss function of the pouch are pouch–anal anastomosis leakage, abscess formation, and pelvic sepsis in the early period and chronic pouchitis in the late period (4). The most frequent long-term complication is ileal pouchitis, the incidence of which is 23–46% in the 10 years after anal anastomosis surgery (3). The most reliable method of diagnosis is endoscopy. In patients who respond to antibiotics, metronidazole (15–20 mg/kg /day) and ciprofloxacin (1000 mg/day) treatment is administered for two weeks. A rare complication of ileal pouch–anal anastomosis surgery is ileal-pouch volvulus, a few cases of which have been reported in the literature (5). Early diagnosis and treatment of ileal-pouch volvulus is important for prevention of pouch necrosis and perforation.

Several cases of ileal J-pouch perforation, a rare but serious complication (5), have been reported in the literature (6). Among the causes of J-pouch perforation, Aouthmany and Horattas reported uterine adhesion in pregnant patients (7), Lontoft reported Salmonella Typhimurium infection (8), and Hsu reported blunt abdominal trauma (9). Shapiro also reported observation of two cases of ileoanal J-pouch perforation subsequent to intake of high-calorie and high-fiber foods (10). In a series of 1,005 patients who had undergone total proctocolectomy, Fazio et al. reported an incidence of para-pouchal abscess and peritonitis of 3.6% but no cases of pouch perforation (3). In the treatment of Ileal J-pouch perforation, defects are repaired primarily or resection is performed with the aid of a blind end stapler. If the pouch requires re-establishment, the distal terminal ileum is used.

The increase in pressure that occurs during defecation may cause blind end expansion. Furthermore, performance of the side-to-side anastomosis technique can lead to intestinal expansion while creating the J-pouch (6). Ileal J-pouch enlargement and thinning of the intestinal wall are considered the main causes of perforation. Therefore, due to the risk of perforation, cases characterized by both pouch dilation and pouchitis should be followed closely. In the case examined here, the ileal J-pouch blind end was 12 cm wide and the wall was too thin. The approach taken here was repairing the J-pouch ileal perforation primarily, performing ileostomy, and treating the pouchitis with antibiotics. Subsequent follow-up of the patient endoscopically revealed normal results at six months surgery.

The J-pouch is the most commonly used type of pouch and J-pouch ileoanal anastomosis the most commonly performed type of procedure, as it yields functional outcomes similar to those of other types of procedures while being much simpler to perform. While there is no difference in terms of postoperative complications among the types of pouches, frequency of defecation and need of antidiarrheal drug use is found to be higher in J-pouch patients (11). However, pouch dysfunction and pouchitis have been reported as late complications in patients who have undergone J-pouch ileoanal anastomosis, and J-pouch perforation has been reported in a very few cases. Type of anastomosis, technique, and length of left blind end may be important factors in the occurrence of these complications, which can be prevented with annual endoscopic examination. Physicians should consider pouch perforation in patient with history of total proctocolectomy and ileal pouch–anal anastomosis who admitted to hospital with the signs and symptoms of acute abdomen.

Conflict of interest

The authors declare that they have no conflict of interest.

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