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CASE REPORT

Jejunoileal Diverticulosis has Heterogeneous Pathophysiology and Requires Special Consideration During Treatment

Jejunoileal Divertiküllerin Patofizyolojisi Değişkendir ve Müdahalesi Özel Dikkat Gerektirir

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ABSTRACT

Jejunoileal diverticulosis (JID) is a rare condition with a reported incidence lower than 0.1%. The clinical presentation of JID varies widely; the incidence of complications requiring surgical intervention is reported as 10%. Surgery is the definitive treatment for JID and can be considered to improve the patient's quality of life and to prevent further severe symptoms. The first patient was a 77-year-old male with a history of JID that had caused intermittent abdominal pain for the last year. He underwent laparoscopic surgery without segmental resection, however, symptoms recurred and he underwent definitive robotic small bowel resection. Pathology revealed JID with true diverticula. The second patient was a 72-year-old male who presented with rectal bleeding that caused hypotension. Jejunostomy was required initially and definitive open surgery was performed later to resect the bowel segment affected by JID. Pathology showed pseudodiverticular JID. JID patients may present with a pseudodiverticulum or a true diverticulum, with severe or mild symptoms and with perforation or minimal inflammation. Physicians treating this heterogeneous disease need to know the complex underlying mechanisms as well as the multiple management options. We share our experience with two distinct cases and discuss the presentation and management approaches for JID to give an inclusive picture of the disease.

Keywords: Jejunoileal, diverticulosis, robotic surgery

ÖZ

Jejunoileal divertikülozis (JID) az görülen bir durumdur ve bildirilmiş insidansı %0,1'in altındadır. Hastaların klinik tablosu değişken olmakla birlikte, cerrahi müdahale gerektiren komplikasyonların sıklığı %10 olarak bildirilmiştir. JID kesin tedavisi cerrahidir; hastaların hayat kalitesini artırmak ve ileride daha ağır semptomlar ile başvurmalarını önlemek için düşünülmelidir. İlk olgu bilinen JID geçmişi olup, son 1 yılda aralıklı karın ağrısı şikayetleri oluşturan 77 yaşında bir erkek hastadır. Segmental rezeksiyon yapılmadan laparoskopik yolla ameliyat edilmiş olmasına rağmen semptomları tekrarlamış ve robotik ince barsak rezeksiyonu uygulanmıştır. Patoloji sonuçlarına göre gerçek divertikülleri olan JID bildirilmiştir. İkinci olgu hipotansiyone yol açan rektal kanama ile başvuran 72 yaşında bir erkek hastadır. Başlangıçtaki müdahelesinde jejunostomi açılmıştır ve daha sonra JID ile etkilenen bağırsak segmentini çıkarmak için açık cerrahi uygulanmıştır. Bu hastanın patoloji sonuçları pseudo-divertikül ile uyumlu bulunmuştur. JID hastaları pseudo-divertikül ya da gerçek divertikül şeklinde, şiddetli veya hafif semptomlarla ve perforasyon ya da minimal enflamasyon ile başvurabilirler. Bu değişken hastalığı tedavi edecek olan hekimler, hastalığın gelişiminin altında yatan kompleks mekanizmaları ve tedavisi için kullanılabilecek birden çok seçeneğini bilmelidirler. Biz bu birbirinden farklı iki olgu ile, hastalığın kliniğe geliş tabloları ve tedavi yaklaşımları ile ilgili tecrübelerimizi paylaşıyoruz.

Anahtar Kelimeler: Jejunoileal, divertikül, robotik cerrahi

Introduction

Jejunoileal diverticulosis (JID) is a rare condition with a reported incidence lower than 0.1%.[1,2,3] Majority of patients with the disease stay asymptomatic but complications of JID can be life threatening. Clinical presentation of JID varies from minimally symptomatic to severely symptomatic with

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symptoms such as chronic pain, malabsorption, perforation, gastrointestinal (GI) bleeding and intestinal obstruction.[4] Exact pathophysiology of JID remains unclear. While an acquired pseudodiverticular type of JID is commonly described, reports of a true diverticular structure in older age is uncommon.[5,6] In this case report, we present two JID cases with different clinical presentations and histopathological characteristics. This report aims to discuss the relationship between pathophysiology and clinical presentation of JID with management strategy for JID.

Case Reports

Case 1

Informed consent was taken from the patient. First patient was a 77 years-old-male with a 1-year history of JID that has caused long standing intermittent abdominal pain. He had several admissions previously due to increased severity of postprandial pain and due to weight loss attributed to his avoidance of eating. Physical examination at most recent admission revealed mild tenderness of the abdomen. The decision was to perform diagnostic laparoscopy. Laparoscopic exploration revealed JID with minimal inflammation. One of the inflamed diverticula on the distal portion was attached to the mesentery of the opposite jejunum and resulted an internal herniation where small intestine segments were internally herniated. The attachment was removed by sharp dissection. Patient was discharged 2 days after surgery without any complications. After 5 weeks, the patient returned with similar symptoms. This time a robotic small intestine resection was planned (da Vinci Surgical System). Robotic resection and intracorporeal anastomosis was performed and the resected specimen was extracted through a mini incision. Postoperative course was uneventful and the patient was discharged on postoperative day 3. Resected specimen was 107 cm long with multiple diverticula (Figure 1). The pathologic evaluation revealed JID with true diverticula and hyperplasia on muscularis mucosa (Figures 2, 3). After the surgery, patient's symptoms improved considerably and he was able to gain weight.

Case 2

Informed consent was taken from the patient. The second patient was a 72 years-old-male, who came to our clinic with rectal bleeding that resulted in hypotension. Initially, he had hemoglobin level of 7.3 and was treated in intensive care unit. In physical examination, the abdomen was distended and rebound tenderness was positive. Chest X-ray revealed suspicion of free air under right hemidiaphragm and computed tomography (CT) was ordered. CT results confirmed presence of free air in the abdomen and immediate laparotomy was planned for the patient. Upon exploration;

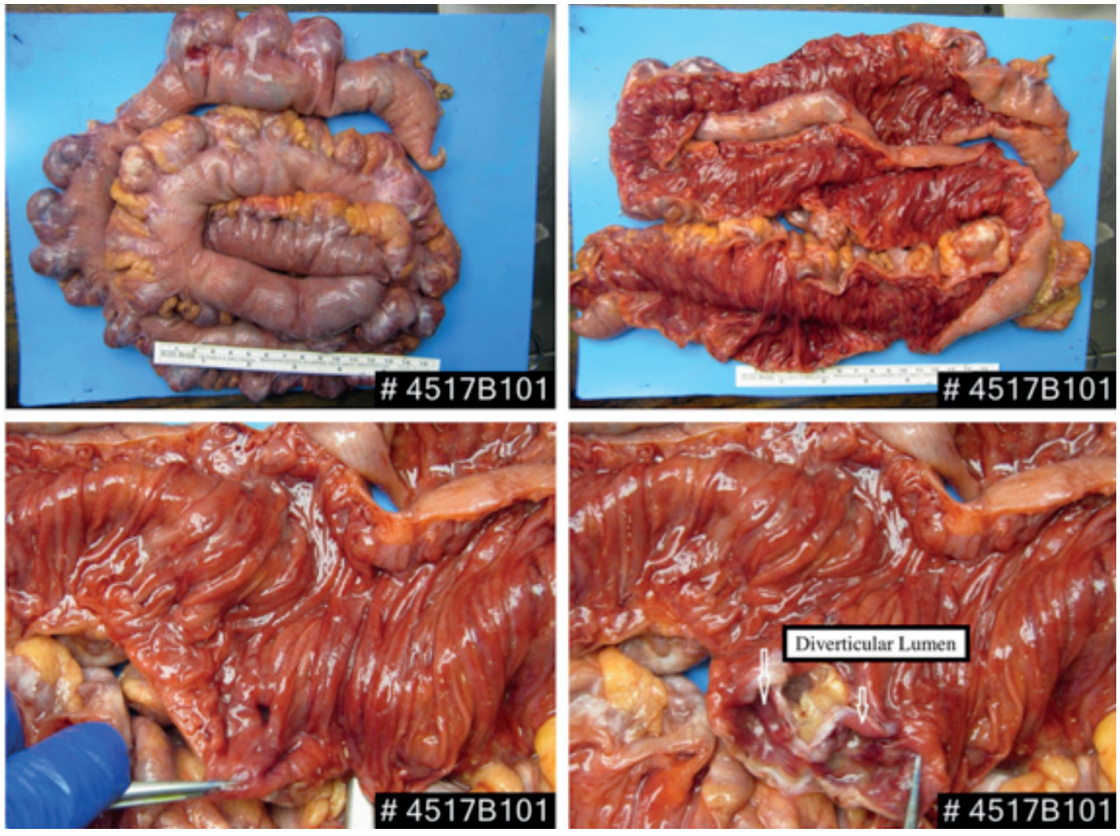

Figure 1. Gross pictures of the resected small bowel specimen on case 1, presenting multiple diverticula

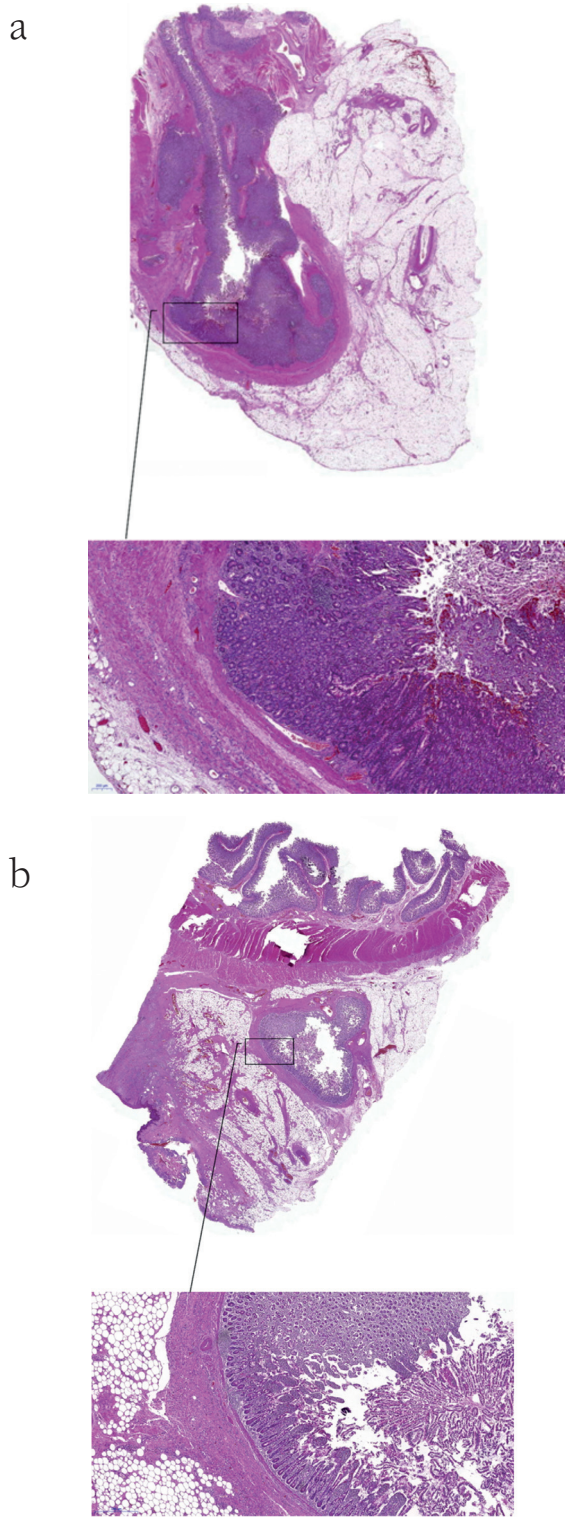

Figure 2. Comparison of diverticula walls of both cases; a) Diverticulum including all layers (mucosa, muscularis mucosa, submucosa, muscularis propria, serosa) of intestinal wall in case 1, b) Diverticulum including only mucosa and submucosa in case 2

jejunal diverticular perforation was observed 20 cm distal to the Treitz's ligament (Figure 4) and a jejunostomy was performed at the level perforation. Postoperatively total parenteral and enteral nutrition support was required due to high jejunostomy output. Six weeks after the initial surgery, definitive open surgery was performed to take down the jejunostomy and resect the affected JID segment. The patient was discharged on postoperative day 10. Resected specimen was 38 cm long with multiple diverticula and diverticulitis. Pathology showed the JID consisted of mucosa and submucosa only without the muscularis propria layer (Figure 2).

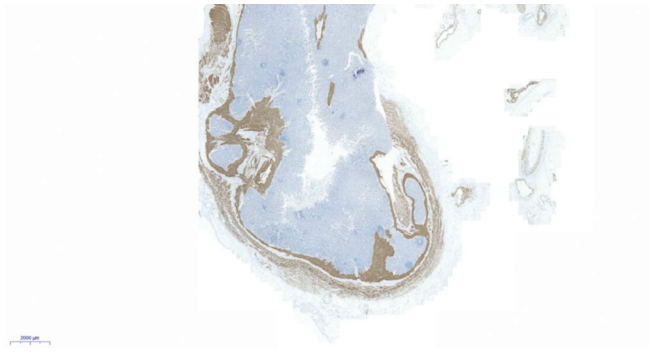

Figure 3. Diverticulum of the case 1 including all layers of the intestinal wall. Conspicuous hypertrophy of the muscularis mucosa is noticed (desmin x0.6)

Discussion

JID is a condition with unclear and multifactorial pathogenesis however, it is generally considered to be of pulsion type where areas of weaknesses on the mesenteric site of the bowel wall result in herniation of mucosa and submucosa, hence the name acquired pseudodiverticula.[6,7] In contrast, there are infrequent cases with a true diverticulum consisting of all layers of the bowel wall.[5,8] It is not possible to make a prediction on the structure of JID on the basis of symptoms of only 2 cases, however, based on our limited experience; the histopathological type might be related to the clinical presentation of JID and may possibly influence the management strategy. Our patient with true diverticular structure, had long standing postprandial abdominal pain but disease never progressed to cause a severe clinical condition. However, for the second patient the pseudodiverticular type JID perforation resulted in a relatively short period of symptomatic disease followed by the development of perforation requiring emergent surgery. Basically, a true diverticulum having all components of the jejunal wall, is expected to be more durable than an acquired jejunoileal pseudodiverticulum which has only mucosa, submucosa, and occasionally a thin layer of serosa without muscle (Figure 2b).[4] Clinical classification of the diverticula can be done as follows; asymptomatic group with the highest incidence of 60%,[8] chronic pain and malabsorption group (also called as minimally symptomatic

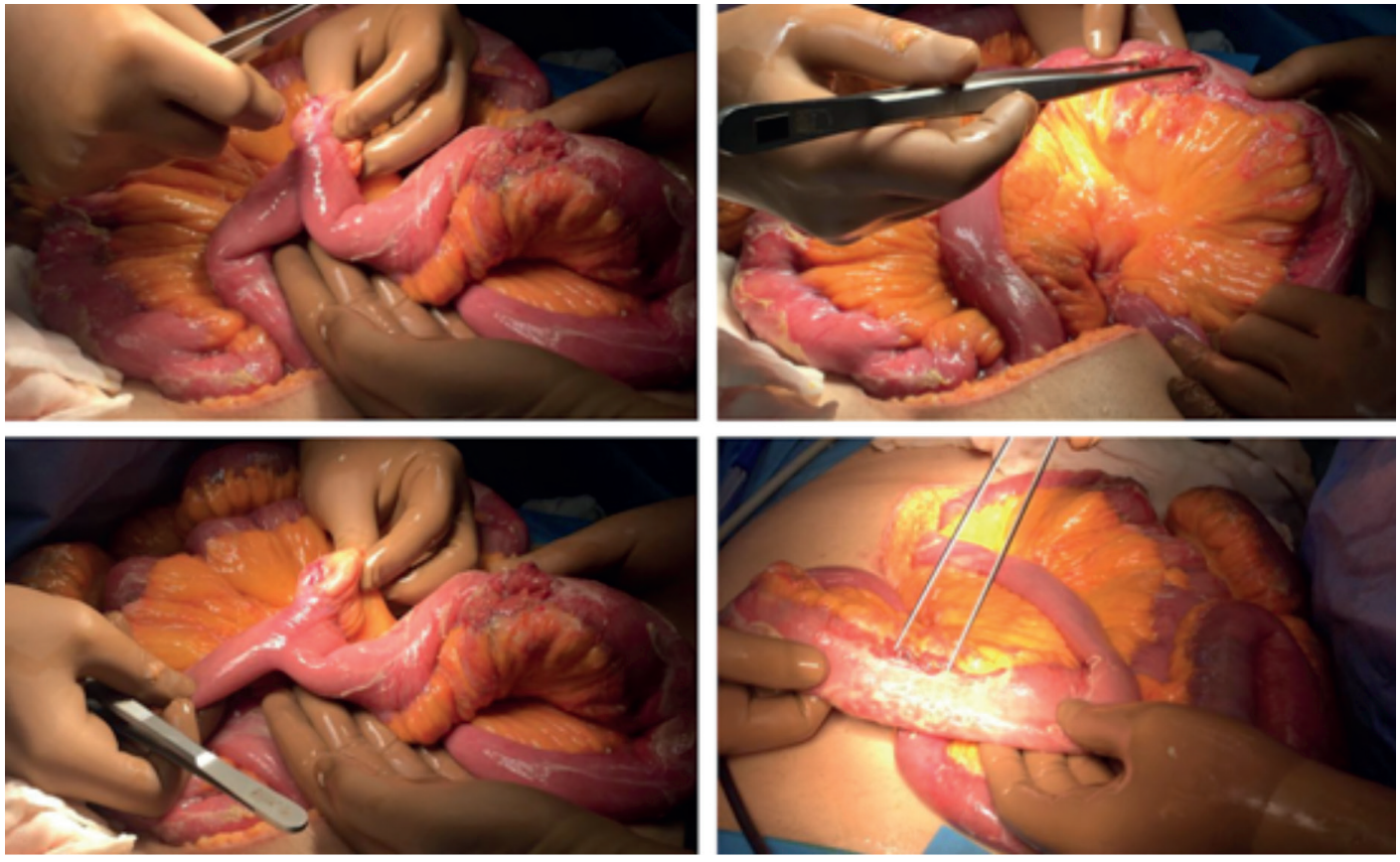

Figure 4. Intraoperative pictures of the perforated diverticulum on case 2

group) and acute complications group.[9] Acute complications may cause considerable levels of morbidity and mortality if there is diverticulitis, perforation, GI bleeding or intestinal obstruction.[5,7] The most common complication is diverticulitis occurring in 2.3-6.4% of all JID patients. It has a high mortality of 21-40% when perforated.[2,10] Management of the perforated small bowel diverticula can be done medically with bowel rest and antibiotics[11] for localized peritonitis and mild abdominal symptoms. In generalized peritonitis, treatment of choice is laparotomy and resection of the small bowel segment.[4,6,12] Our second case had perforated JID with generalized peritonitis and required a diverting stoma at the time of index surgery. We performed a diversion to save the patient's life and electively resected diseased part of the jejunum with jejunojejunal anastomosis without diversion. Among complications of JID, mechanical obstruction is the most frequent complication requiring surgery with an incidence of 2.3% to 4.6% of JID.[1,7,13] Mechanisms of obstruction include adhesion, intussusception, volvulus. Our first case developed an obstruction due to adhesion between diverticula and abdominal wall. Since the histopathologic type of JID cannot be determined preoperatively, clinical signs and symptoms should be carefully evaluated together with the condition of patient at the time of admission or at the first visit. As mentioned above, conservative treatment can be an option when there are no life-threatening complications, and, although overall incidence of complications requiring surgery is reported as 10%, operative approach is still the definitive treatment and can be preferred to improve patients' quality of life and to prevent more severe symptoms from developing.[3,7] Additionally, surgical approach can be considered even for patients with chronic symptoms.[14] Laparoscopic or robotic exploration and evaluation is as effective as open surgery especially in elective conditions. Minimally invasive surgery facilitates recovery and provides better outcomes of JID treatment with less risks.[15] Robotic approach facilitates intracorporeal anastomosis and transection of bowel whereas those operative steps can be more challenging during laparoscopy. We did not have any postoperative complications after robotic removal of the diseased jejunum in our case. Satisfactory operative outcomes and postoperative recovery were facilitated with the help of advanced minimally invasive techniques. Improved outcomes, feasibility, and both diagnostic and therapeutic features of the minimally invasive techniques, reduce the threshold for operating patients with vague symptoms for JID. We cannot suggest structure of JID as

a predictor of clinical course or as a parameter in planning the treatment strategy. However, our report shows that JID has heterogeneous pathophysiology and requires special attention care.

Ethics

Informed Consent: Informed consent was taken from the patient.

Peer-review: External and internal peer-reviewed.

Authorship Contributions

Surgical and Medical Practices: E.A., İ.E.B., İ.A.B., Concept: E.A., İ.E.B., İ.A.B., Design: E.A., F.B.B., T.K.Y., E.A., İ.E.B., İ.A.B., Data Collection or Processing: F.B.B., T.K.Y., İ.A.B., Analysis or Interpretation: E.A., F.B.B., T.K.Y., Literature Search: F.B.B., T.K.Y., Writing: F.B.B., T.K.Y., E.A.

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