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Unusual Differential Disease Entity of Right Lower Abdominal Pain, Ileal Diverticulitis Perforation: A Report of Three Cases

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Introduction: Small bowel diverticulum is a rare disease entity, and most cases are asymptomatic. However, diverticulitis can cause acute abdomen conditions like colonic diverticulitis. Depending on the location, various abdominal symptoms may ensue. Clinical manifestations of ileal diverticulitis may be similar to those of appendicitis and or colonic diverticulitis. Thus, making a diagnosis based on a physical examination alone may be challenging. Even though imaging techniques such as computed tomography (CT) may provide clues for a definite diagnosis, the majority of cases are confirmed through operation. Here, we present 3 cases of ileal diverticulitis perforation, which were not diagnosed preoperatively.

Case presentation: A 71-year-old man, a 77-year-old woman, and a 78-year-old woman presented with abdominal pain. All 3 patients showed local peritoneal irritation signs in the right lower quadrant. Appendicitis or colonic diverticulitis was suspected, but CT scan results revealed free air and mesenteric fat infiltration around the terminal ileum, suggesting ileal perforation. During the operation in each case, terminal ileal perforation was confirmed, and ileocecal resection was performed. Subsequently, histologic examination revealed ileal diverticulitis perforation. Although the postoperative course was uneventful in the first and second patient, the third patient died of sepsis resulting from anastomosis leakage.

Conclusion: These cases put forth unusual causes of right lower quadrant pain, which show physical findings similar to those of diverse inflammatory disease. Awareness of this

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disease is beneficial for making a differential diagnosis based on CT findings and eliciting prompt surgical management.

Key words: Diverticulitis – Perforation – Small bowel – Ileum – Differential diagnosis

Ileal diverticulitis is one of those rare disease entities, with an incidence of less than 1.5%; however, it is the most common complication of small diverticula disease.[1] Progression of the inflammatory process in the diverticula disease typically results in bleeding, obstruction, and perforation.[2,3] Unlike colonic diverticulitis perforation, in ileal diverticulitis, the peritoneal cavity is soiled by the small bowel content, with subsequent less fecal contamination. Thus, operative management may be performed without fecal diversion, and the prognosis of ileal diverticulitis is better than that of colonic diverticulitis perforation. However, even though the findings of perforated ileal diverticulitis in imaging studies have been reported, since the incidence of this condition is quite low and the findings of physical examination are similar to those of inflammatory diseases that present in the right lower quadrant abdomen, such as appendicitis, cecal diverticulitis and Crohn's disease, a definitive preoperative diagnosis is rarely made, particularly in the emergency room. This may ultimately result in a delay of treatment, thus leading to morbidity and mortality.[4–6] Here, we present 3 cases of ileal diverticulitis perforation, which were confirmed through operative findings and histologic examination.

Case Presentation 1

A 71-year-old man presented to the emergency department with right side abdominal pain and distension beginning three days prior. He had been diagnosed with alcoholic liver cirrhosis 10 years ago with ongoing regular follow-up occurring at the department of gastroenterology, and a history of hypertension. Although he denied nausea, vomiting and or dyspepsia, his abdominal pain had worsened over the time and a fever had developed prior to the visit. Initial vital signs at presentation to the emergency department were as follows: a temperature of 38.2°C, blood pressure of 155/85 mmHg, a pulse rate of 98 beats/min and a respiratory rate of 23 breaths/min. The patient showed acute ill appearance and, on physical examination, his abdomen was slightly distended, and even though signs of generalized peritoneal irritation signs were not apparent, local tenderness and rebound tenderness were evident in the right lower quadrant abdomen. Laboratory test results were within the normal limits, including liver function test, except for a white blood cell count (WBC) of 16,000/μL. Based upon both the physical examination results and laboratory data, perforated appendicitis or perforated diverticulitis was suspected. However, aside from liver cirrhosis with fatty liver and esophageal varix, a CT scan also revealed diffuse edematous wall thickening of the ascending colon and ileal loop, mesentery haziness, peritoneal wall thickening, fluid collection, and free air around the terminal ileum (Fig. 1). Under the impression of a small bowel perforation caused by inflammatory bowel disease such as Crohn's disease, emergency *laparotomy* operation was performed. During the operation, purulent fluid was collected in the paracolic gutter and pelvis, and about a 1×0.5 cm measured perforation site was found approximately 10 cm from the ileocecal valve on the mesenteric side, which was sealed with omentum. Following irrigation of the abdominal cavity and adhesiolysis of the omentum, ileocecal resection with *side-by-side primary anastomosis using linear staplers* was performed. A subsequent histologic examination showed diverticulitis with perforation and abscess formation at the terminal ileum. Even though wound infection was developed postoperatively, the patient was discharged on the 17th postoperative day with conservative wound management.

Case Presentation 2

A 77-year-old woman presented to the emergency department with diffuse abdominal pain beginning 3 days prior. She was hospitalized for dementia 6 months prior to the current visit in a local clinic with bed-ridden status, and had a 30-year history of hypertension and diabetes. Although conservative management for the pain was initially attempted, the pain was ultimately aggravated without improvement over the time. On physical examination, she appeared chronically ill and had a temperature of 38.5°C, blood pressure of 140/70 mmHg, a pulse rate

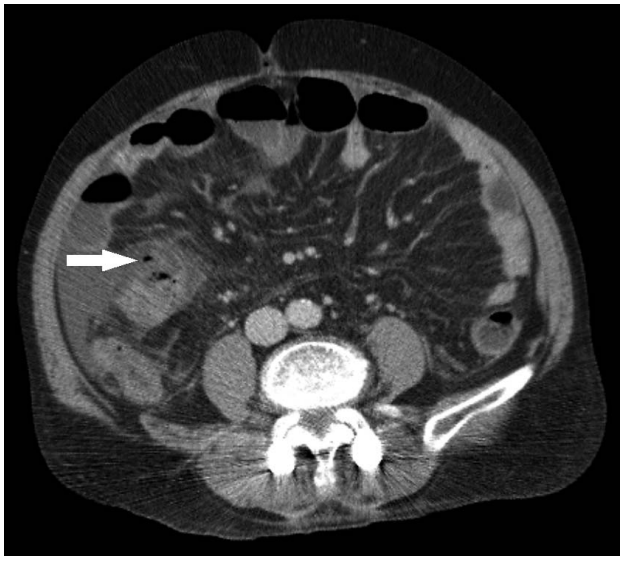

Fig. 1 CT of case 1 indicates diffuse edematous wall thickening of the ascending colon and ileum, which is accompanied by diffuse mesentery haziness, fluid collection, fat infiltration, and free air (arrow).

of 99 beats/min, and a respiratory rate of 22 breaths/min. Her abdomen was soft and flat, but tenderness, rebound tenderness, and muscle guarding were apparent over the right lower quadrant abdomen. Laboratory test results were within the normal limits, except for a white blood cell count (WBC) of 12,000/μL. Multifocal air density, fluid collection, mesenteric fat infiltration around terminal ileum with wall thickening, and multiple diverticulum at the proximal colon and cecum were noted on the CT scan (Fig. 2). Under the impression of colonic diverticulitis perforation, an emergency *laparotomy* operation was

Fig. 2 CT of case 2 reveals multifocal air density with fluid collection and fat infiltration around the terminal ileum. Diverticulum of the cecum is noted (arrow).

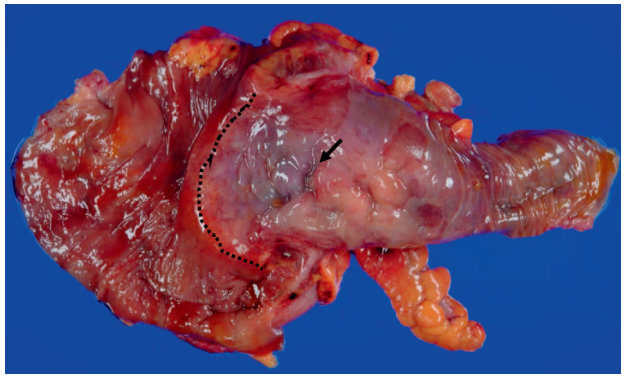

Fig. 3 Gross photograph of ileocecectomy of case 2. An ostium of diverticulum (arrow) is seen in the terminal ileum and the adjacent mucosa showed inflammation (dotted line, ileocecal valve).

performed. However, during the operation, although multiple diverticulum was noted on the ascending colon and cecum, there was no evidence of inflammation and/or perforation. Interestingly, however, near to the ileocecal valve on the mesenteric side of terminal ileum, a pinpoint perforation site was detected. Ileocecal resection with *side-by-side primary anastomosis using linear staplers* was performed. A pathologic report showed diverticulitis with perforation at terminal ileum and diverticulosis on the cecum (Figs. 3, 4). The postoperative course was uneventful, and the patient was discharged on the 8th postoperative day.

Case Presentation 3

A 78-year-old woman presented to the emergency department with right lower quadrant abdominal pain beginning one days prior. She had no significant medical history, except for well-controlled diabetes mellitus and hypertension, which were both diagnosed 10 years ago. The patient visited an out-patient department of orthopedic surgery due to back pain 1 day prior to presenting to the emergency department. After returning home, however, abdominal pain developed and worsen over time. Upon physical examination, the patient demonstrated an acute ill appearance. Her vital signs at the emergency room are as follows: a temperature of 37.8C, blood pressure of 145/70 mmHg, a pulse rate of 95 beats/min and a respiratory rate of 22 breaths/min. Her abdomen was soft and flat, but tenderness and rebound tenderness were evident on the right lower quadrant. Laboratory test results were within the normal limits, except for a white blood cell count

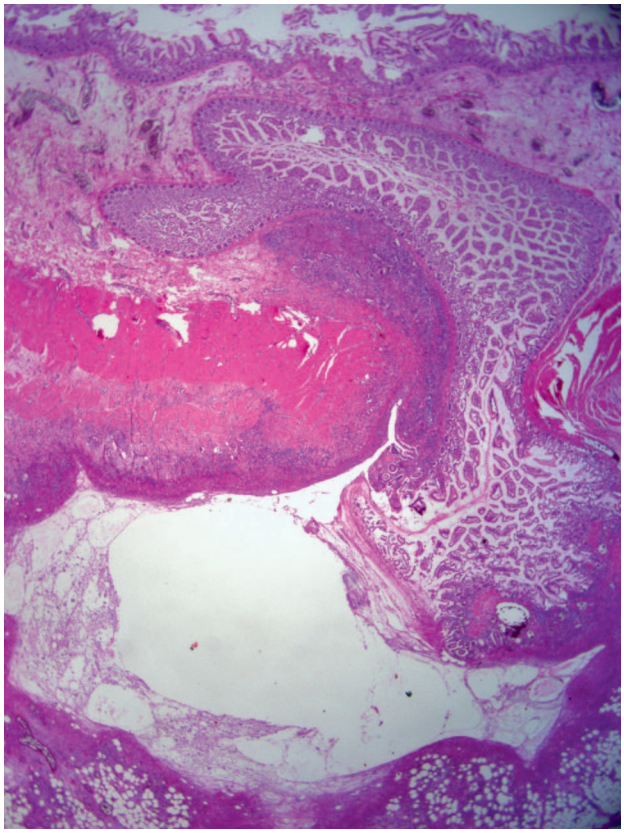

Fig. 4 The ileal mucosa of case 2 herninates through the muscularis propria and acute suppurative diverticulitis is seen (Hematoxylin and eosin × 10).

(WBC) of 15,300/μL. Although there were no remarkable findings with respect to a simple view of the abdomen, a CT scan revealed pericolic fat infiltration, peritoneal thickening, and air loculation in the right lower quadrant area (Fig. 5). Under the impression of perforation in terminal ileum and an ascending colon or cecum by unknown cause, an emergency *laparotomy* operation was performed. During the operation, the appendix was normal, but along the minimal amount of purulent fluid collection at the right paracolic gutter, a 0.5 × 0.5 cm measured perforation on the mesenteric border, 20 cm from the ileocecal valve, was identified. Ileocecal resection with *side-by-side primary anastomosis using linear staplers* was performed. Pathologic results were consistent with ileal diverticulitis perforation. However, anastomosis site leakage was detected on the eighth postoperative day. Reoperation was scheduled, but the patient's guardians refused further treatment. Thus, the patient died of multi-organ failure by sepsis on the 15th postoperative day.

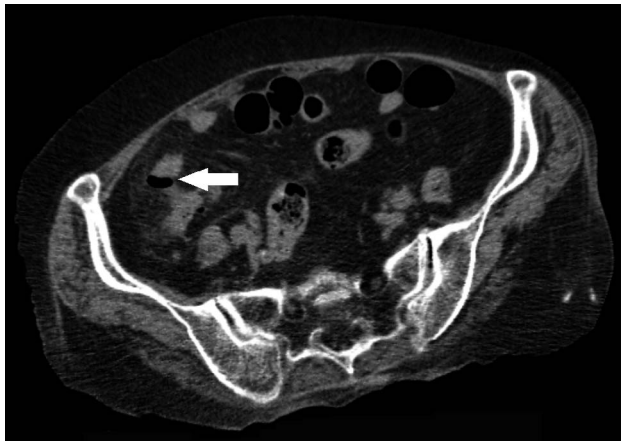

Fig. 5 CT of case 3 indicates pericolic fat infiltration and air loculation in right low quadrant (arrow).

Discussion

Right lower quadrant abdominal pain is commonly encountered in the emergency room. Depending on the findings of physical examination, however, various diseases can be considered as a differential diagnosis, including appendicitis, mesenteric lymphadenitis, diverticulitis, pelvic inflammatory disease and or inflammatory bowel disease, in which local peritoneal irritation signs including tenderness and rebound tenderness may present, indicating an ongoing inflammatory process in the abdomen. Thus, based on physical findings, along with laboratory results, imaging studies are performed for the definite diagnosis and treatment strategy. The diagnosis of common disease entities such as appendicitis and diverticulitis presenting with right lower quadrant pain can typically be easily diagnosed through physical findings and imaging studies; however, rarer disease entities like the present cases may be more challenging to determine when making a definite diagnosis. In such cases, definite diagnosis is made during operation or after histologic evaluation. Ileal diverticulitis is one of these rare disease entities, which similarly presents with right lower quadrant local inflammation.[4,5]

Even though small bowel diverticulum is rare, it can be observed throughout the whole small bowel ranging from the duodenum to the ileum. The duodenum is the most common site, and the incidence of jejunal diverticulum is much higher than that of ileal diverticulum. Unlike true diverticulum, so-called Meckel's diverticulum, however, most of the small bowel diverticula are false diverticula, which consist of mucosa, submucosa,

and serosa without muscle layer, and are formed at the weak, mesenteric side, where blood vessels penetrate. Thereby, since the jejunum has larger penetrating vessels comparing with those of ileum, the incidence of jejunal diverticulum is more common than that of ileum diverticulum.[7,8]

Clinically, small bowel diverticulum is more common in males than in females, and is frequently combined with colonic diverticulum.[2,9] Even though the pathogenesis has not been well established, a lack of dietary fiber resulting in a pseudo-obstruction and bowel movement abnormalities caused by the smooth muscle or mesenteric plexus malfunction have been suggested as possible mechanisms, much like colonic diverticulum.[10–12]

The rate of complications of ileal diverticulum is approximately 10%, which includes diverticulitis, hemorrhage, obstruction, abscess formation, and perforation. Of them, diverticulitis may be the most common complication.[2,3,9] Even though the risk of diverticulitis followed by perforation is not known, 1 study reported that 2 of 9 diverticulitis patients were diagnosed as having ileal diverticulitis perforation. In the study, the average age of the diverticulitis patients was 43.7 years, but the patients with perforation were old-aged (71 and 67 years old, respectively).[2] In the present cases, although only 1 patient had concurrent colonic diverticula, all 3 patients were older than 70 years of age.

In general, ileal diverticulum does not cause symptoms like colonic diverticulum, but inflammation of the diverticulum can cause acute abdomen. Since the ileum, particularly the terminal ileum, is close to the appendix and ascending colon/cecum, ileal diverticulitis can give rise to symptoms and signs similar to those of appendicitis, colonic diverticulitis, pelvic inflammatory disease, and or Crohn's disease, which are differential diagnoses in patients with right lower quadrant abdominal pain.[4,5] So, it is difficult to diagnose ileal diverticulitis perforation based upon a physical examination and laboratory testing completed before imaging studies because it is not only a rare disease, but also, its inflammatory process occurs in the same region of the abdominal cavity as in other conditions. Moreover, in the Asian population, right side colonic diverticulitis, such as cecal diverticulitis, prevails over left side diverticulitis, and differential diagnosis between appendicitis and cecal diverticulitis in such a case through just physical examination is difficult.[13] Thus, ileal diverticulitis and perforation may be included in the differential diagnosis, along with appendicitis and diverticulitis for patients with right low quadrant pain. In the present cases, all 3 patients showed peritoneal irritation signs in the right lower quadrant, thereby, the presumptive preoperative diagnoses were appendicitis or diverticulitis with/without perforation.

The diagnosis of ileal diverticulitis may be made based on imaging studies such as computed tomography (CT), sonography, and barium study.[2,3,6] In the emergency room, sonography is readily accessible, but it is practitioner-dependent, and bowel gas may hinder the successful evaluation of the diverticulum. Barium study is another option for detecting diverticulum; however, it is usually not available in the emergency room and may be contraindicated in cases of perforation. So, CT may be the choice for imaging technique for examining ileal diverticulitis perforation in the emergency situation.[2,6] Ileal wall thickening, around mesenteric fat infiltration, free air, and diverticulum are all characteristic findings that can be identified, as seen in the present cases. However, even though these imaging studies may provide clues for the diagnosis, a definite diagnosis of ileal diverticulitis perforation is most often made in the operation room, or even after histologic report.[5] In the present cases, although the CT findings were suggestive of perforation of the ileum and the perforation site was identified during the operation, a definite diagnosis was made through histologic report. This may be attributed to the rarity of the cases and lack of experience regarding such a disease.

Non-operative management methods including bowel resting and intravenous antibiotics are treatments of choice in ileal diverticulitis, as like in colonic diverticulitis.[2,4] Even though some degree of success of conservative management has been reported in patients with perforation, surgical treatment is usually necessary in order to prevent disease progression that can lead to sepsis.[2] Surgical options are adopted according to the patient's condition and the degree of peritoneal soling, but, unlike in colonic perforation, diversion stoma seldom, if ever, is performed. Resection and primary anastomosis, such as ileocecal resection and right hemicolectomy, would be enough of a surgical option in cases of ileal diverticulitis perforation. Wedge resection of the perforated diverticulum may be another surgical option because it requires a short operative time. However, sometimes, bowel edema and inflammatory changes in the ileum may make it difficult to perform wedge resection and primary repair. Thus, although standard surgical options have not been established due to the rarity of the condition, surgical

modality should be selected case by case. The mortality associated with small bowel perforation ranges from 12% to 19%, and high Mannheim Peritonitis Index (MPI) score and preoperative hypotension have been reported as risk factors for mortality.[3,14] The MPI score reflects the degree of peritonitis, which is calculated using several factors such as age and preoperative duration of peritonitis. In the present cases, we experienced 1 mortality case. Although the mortality resulted from untreated anastomosis leakage, old age and peritonitis over 24 hours may contribute as a precipitating factor.

Conclusions

Physical examination and laboratory results can provide critical clues towards diagnosis in the emergency room. However, making presumptive diagnoses using such information is challenging in patients presenting with right lower quadrant abdominal pain because there are a wide variety of possible inflammatory diseases. Even though ileal diverticulitis perforation is a rare disease entity and its diagnosis is mainly made during the operation, it should still be considered as a differential diagnosis in patients with right lower quadrant pain because suspicion aroused by CT findings, followed by prompt surgical management, is crucial for the prevention of morbidity and mortality.

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