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Case Report



A Case of Transverse Colon Located Pneumatosis Cystoides Intestinalis Requiring Surgical Resection

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Abstract

Pneumatosis cystoides intestinalis (PCI) is a rare disease characterised by numerous gas-filled cysts in the intestinal wall. This case report is aimed to present the diagnosis and treatment process of a case of PCI treated with intestinal resection. A 56-year-old female was admitted to the emergency department of a tertiary health centre with complaints of abdominal pain and vomiting for about two days. In the abdominal examination of the patient, there was widespread tenderness and defence in all abdominal quadrants with a rebound. Laboratory parameters of the patient were unremarkable. The plain abdominal X-ray observed no free air in the right subdiaphragmatic area. Computed tomography showed dilatation, increased wall thickness and multiple free airs in the small bowel loops, predominantly localised in the left upper quadrant. The patient underwent a diagnostic laparotomy. Diffuse air bubbles were observed along the transverse colon wall at laparotomy. The patient underwent transverse colon resection and colocolic anastomosis with a linear stapler. The patient was discharged on the 7th postoperative day. The pathological evaluation of the surgical specimen was suitable with PCI.

Keywords: Pneumatosis cystoides intestinalis, intestinal perforation, large bowel resection

INTRODUCTION

Pneumatosis cystoides intestinalis (PCI), first described by the pathologist DuVernoi, is a rare disease characterised by numerous gas-filled cysts in the intestinal wall (1). The condition, classified in two forms as idiopathic and secondary, may present different clinical findings (2). Air cysts in the idiopathic or primary form are located in the mucosa and submucosa. The air cysts are primarily found in the intestinal wall in the secondary form. The most common form of this disease is the secondary form. There are uncertain and varied hypotheses for the aetiology of PCI. It possibly occurs due to an underlying cause such as trauma, inflammation, drug use, immunosuppression, neoplasia, and autoimmune or pulmonary diseases (3).

PCI clinics range from asymptomatic disease to severe peritoneal disease. Imaging tools such as plain radiography and computed tomography, endoscopy, surgery and sometimes pathological examinations are used in diagnosis. While hyperbaric oxygen therapy, antibiotics and bowel rest are used to treat asymptomatic patients, surgical treatment is recommended in patients with peritonitis symptoms (4). In surgical treatment, a biopsy from pathologic areas should be taken. Resection is not required in cases without significant perforation and gastrointestinal system content in the abdominal cavity. In contrast, resection with or without anastomosis (with a diverting ostomy) should be performed in cases with suspicious or obvious perforation and gastrointestinal tract infection, depending on the infection amount of the abdominal cavity (5).

This case report is aimed to present the diagnosis and treatment process of a case of pneumatosis cystoides intestinalis treated with intestinal resection.

CASE REPORT

A 56-year-old female was admitted to the emergency department of Erzurum Regional Education and Research Hospital with complaints of abdominal pain and vomiting for about two days in May 2022. She did not have a similar episode of abdominal pain before. She had no other disease, and she had only a history of laparoscopic cholecystectomy ten years ago.

CITATION

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At the time of admission to the hospital, the patient's arterial blood pressure was 142/80 mmHg, oxygen saturation on room air was 95%, pulse rate was 105 beats/ min, and body temperature was 37.6°C. In the abdominal examination of the patient, there was widespread tenderness and defence in all abdominal quadrants with a rebound. Other system examinations, including rectal examination, were normal.

Laboratory parameters of the patient were unremarkable. The plain abdominal X-ray observed no free air in the right subdiaphragmatic area. Since she had severe abdominal pain, abdominal computed tomography (CT) with contrast was performed to evaluate the patient. Computed tomography showed dilatation and increased wall thickness in the small bowel loops, predominantly localised in the left upper quadrant (Figure 1). In addition, there were numerous air bubbles on the transverse colon wall (Figure 2).



Figure 1. On computed tomography scan, yellow arrows indicate dilated intestinal segments



Figure 2. A yellow arrow indicates the free air inside the transverse colon wall on a computed tomography scan

The patient underwent a diagnostic laparotomy. At laparotomy, diffuse air bubbles were observed along the transverse colon wall (Figure 3). No additional gastrointestinal organ perforation was observed. No ischemic bowel loop was observed. Resection anastomosis was planned because the PCI area was extended, and the intact colon wall was low. The patient underwent transverse colon resection with a colocolic anastomosis via a linear stapler. One drain was inserted from the right side to the edge of anastomosis, and the other was inserted into Douglas's pouch. She was followed up in the service during the postoperative period and was started on ceftriaxone 1 gr vial (intravenously every 12 hours) and metronidazole 500 mg/100 ml (intravenously every 8 hours). Oral feeding of the patient, whose complaints regressed in the postoperative period, was opened on the 3rd postoperative day. The drain of the patient who tolerated oral feeding was removed on the 6th postoperative day, the patient's current antibiotherapy was completed for seven days, and the patient was discharged on the 7th postoperative day. The pathological evaluation of the surgical specimen was suitable with PCI.



Figure 3. Perioperative image of the resection material (Yellow arrows indicate multiple air bubbles along the transverse colon wall)

DISCUSSION

Pneumatosis cystoides intestinalis (PCI) is a rare pathology of unknown aetiology, characterised by multiple gas-filled cysts in the intestinal subserosa and submucosa. The cyst content consists of nitrogen and hydrogen and various amounts of oxygen, carbon dioxide, butane, propane, methane, ethane, and argon gases (6). In the autopsy series of 6553 cases performed at Edinburgh Hospital, PCI was detected in only two patients (0.03%) (7). PCI occurs equally in men and women and is most common in the 6th decade (8). In the present case, the patient was a female, and she was 56 years old, which was compatible with the age at which PCI was most common.

Most PCI cases involve the jejunum and ileum; in 6% of the cases, the colon is affected. Many gastrointestinal system diseases include appendicitis, Crohn's disease, pyloric stenosis, ulcerative colitis, diverticular disease, necrotising enterocolitis, gastroduodenal ulcer, and sigmoid volvulus may accompany PCI. In addition, PCI is unknown in cases unrelated to the gastrointestinal system, such as chronic obstructive pulmonary disease, collagen tissue diseases, AIDS, and glucocorticoid use. Pathology accompanying PCI could not be detected in approximately 20% of the cases, and the disease was accepted as primary (9). There was no underlying disease in this case, and the case was evaluated as primary PCI.

Although the pathogenesis of PCI is unknown, some theories have been proposed. As a result of causes that increase intrathoracic pressure such as emphysema, asthma, mechanical ventilation, vomiting and vomiting, the alveoli are fragmented, and the gas content passes from the mediastinum retroperitoneum and mesentery to the intestinal wall (6). In addition, it has been suggested that the disease may also occur due to the gas-forming bacteria (*Clostridium Difficile, Clostridium perfringens*, etc.) releasing hydrogen through carbohydrate fermentation and these gases invading the lymphatic vessels and the intestinal wall (10).

Direct abdominal X-ray and abdominal computed tomography (CT) are valuable imaging modalities in diagnosing PCI. CT is the ideal method for diagnosis, although plain abdominal radiography gives positive findings in 2/3 of the cases (11). However, radiological diagnosis is difficult if the patient has a good prognosis and pneumoperitoneum is not detected. In addition, air appearances outside the intestinal lumen were seen in the vicinity of the small intestine wall, where the distinction between free air and PCI could not be made clear (12). In the present case, there was no free air in the right subdiaphragmatic area. On CT scan, in some small bowel loops, localised dilatation, increased wall thickness and multiple free air densities in the abdomen, especially in the left upper quadrant, were observed.

Since spontaneous remission is observed up to 50% in the treatment of the disease and gas cysts can reoccur after surgery, no specific treatment is applied in asymptomatic cases. Nonoperative oxygen therapy is used first if there is no perforation, peritonitis, or sepsis in symptomatic patients (11). According to a study published by EAST (Eastern Association for the Surgery of Trauma) in 2013, various risk factors have been defined in making the surgical decision (13). Hypotension, use of vasopressor drugs, peritonitis, lactate level above two mmol/L, signs

of acute renal failure, and mechanical respiratory support were accepted risk factors for pathological PCI. Apart from those mentioned in this study, age 60 and over, nausea, bicarbonate level below 20 mmol/L and white blood cell value above 12x109/L are the clinic-laboratory factors. Gas in the portal vein, free air in the abdomen, enlargement of the intestine, and presence of ascites have also been defined as CT factors (13). In surgical treatment, a biopsy from pathologic areas should be taken. Resection is not required in cases without significant perforation and gastrointestinal system content in the abdominal cavity. In contrast, resection with or without anastomosis (with a diverting ostomy) should be performed in cases with suspicious or obvious perforation and gastrointestinal tract infection, depending on the infection amount of the abdominal cavity (5). Segmental resection with anastomosis was performed in the present case due to suspicious colon wall perforation.

CONCLUSION

While pneumatosis intestinalis may develop due to a disease that can be treated on an outpatient basis in some cases, it can sometimes be a sign of conditions that can be fatal if surgical intervention is delayed. It is essential to evaluate this finding with the patient's complaints, history, comorbidities, physical examination findings, and laboratory and other radiological examinations to delay the diagnosis and apply the proper treatment.

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Informed Consent: The family provided consent for publication.

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