Cukurova Medical Journal
ÇUKUROVA ÜNİVERSİTESİ TIP FAKÜLTESİ

Cukurova Med J 2018;43(2):463-466
DOI: 10.17826/cumj.342572

OLGU SUNUMU / CASE REPORT

Pneumatosis cystoides intestinalis after endoscopic balloon dilatation

Endoskopik balon dilatasyonu sonrasında gelişen pnömatozis sistoides intestinalis

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Cukurova Medical Journal 2018;43(2):463-466

Abstract
Pneumatosis cystoides intestinalis (PCI) is a rare disease of unknown etiology that is characterized by subserosal and submucosal gas cysts in the gastrointestinal system. Many gastrointestinal system diseases, such as pyloric stenosis, may be associated with PCI. Cases of PCI secondary to surgery or endoscopic trauma have also been reported. We re ported. Here we present a case of PSI that was detected during surgical treatment with pyloric stenosis and after endoscopic dilatation treatment.
Key words: pneumatosis, cystoides, balloon dilatation, pyloric stenosis

Öz
Pnömotosis sistoides intestinalis (PSİ), etiyolojisi tam olarak bilinmeyen, gastrointestinal sistemde subserozal ve submukozal gaz dolu kistler ile karakterize nadir görülen bir hastalıktır. Pilor stenozu gibi birçok gastrointestinal sistem hastalığı PSİ'ye eşlik edebilir. Ayrıca cerrahi veya endoskopik travmaya sekonder PSİ olguları da bildirilmiştir. Burada pilor stenozu olan ve endoskopik dilatasyon tedavisi sonrasında yapılan cerrahi tedavi esnasında saptanan PSİ olgusu sunulmuştur.
Anahtar kelimeler: pnömatozis, sistoides, balon dilatasyon, pilor stenozu.

INTRODUCTION

Pneumatosis cystoides intestinalis (PCI) is a rare disease of unknown etiology that is characterized by subserosal and submucosal gas cysts in the gastrointestinal system[1]. First described in 1730, it has been given a variety of names such as pneumatosis intestinalis, pneumatosis coli, intestinal lymphocysts and intestinal emphysema[2].

Three theories have been put forward to explain the gas-filled cysts. The mechanical theory suggests that gas from the lumen and the serosa passes through the intestinal wall, the bacterial theory suggests that gases produced by bacteria in the lumen penetrate the intestinal wall through small areas of damaged mucosa, and the biochemical theory suggests that large amounts of gas produced by bacteria infiltrates the lymphatic vessels and the intestinal wall[3,4]. PCI is usually asymptomatic and might resolve on its own if the underlying disease is treated. Treatment options include steroids, elemental diets, hyperbaric oxygen, antibiotics and surgical approaches[3,5,6].

PCI might develop secondary to other clinical conditions (85%), or as a primary condition (15%). While the incidence peaks in the 25-50 age group, it can also occur in the first days of life and in old age[6]. PCI is not a diagnosis, but rather a radiological or explorative finding[8]. The true prevalence of PCI is currently unknown. An autopsy series reported the overall prevalence to be 0.03%[5], another series performed by Theisen reported only 2 cases of PCI among 6553 autopsy patients[9], but many clinicians believe the true prevalence is much higher.

Many gastrointestinal system diseases, such as pyloric stenosis, may be associated with PCI[8,10]. Cases of PCI secondary to surgery or endoscopic trauma have also been reported[8]. In this article, we aimed to present a case of PCI that was discovered during pyloric stenosis surgery.

CASE

The patient was a 62-year-old man with complaints of hematemesis. The man had first presented to the

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Geliş tarihi/Received: 25.04.2017 Kabul tarihi/Accepted: 30.07.2017

emergency department over a year ago. He had received three units of Red Blood Cell suspension after his hemoglobin levels dropped from 12.4 g/dL to 8.6 g/dL. A subsequent esophagogastroduodenoscopy had revealed pyloric stenosis and 2 ulcerated lesions located in a hyperemic area nearly 4cm in size at the prepyloric region. Biopsies were performed, and the lesions were found to be benign. Abdominal CT revealed no pathological findings other than the thickening of the distal esophageal walls and enlargement of the distal esophagus, lobulated appearance of the stomach fundus and corpus, thickening of the rugae, and narrowing of the pylorus. The patient then underwent 3 endoscopic balloon dilation procedures.

The patient complained of dysphagia, inability to lie on his back and abdominal distension, and could not eat solid foods, but only consume liquids. He was later admitted to our clinic for surgery after losing 10 kg weight in one year despite medical treatment.

During exploration, the stomach was found to be extremely dilated (Figure 1), and on the intestines cystic formations limited to the serosa were observed, affecting a 120 cm length of small intestine starting 1 meter after Treitz ligament (Figure 2, 3, 4). In addition, polypoid formations were palpated in the lumen of sigmoid colon. Biopsy samples obtained from the serosa of small intestine and the polypoid formations inside the sigmoid colon were taken to intraoperative pathological assessment. No signs of malignity were found, and the lesions were reported compatible with pneumatosis cystoides intestinalis (PCI). Subsequently, uncut gastrojejunostomy (with a jejunum segment prepared 50 centimeters from Treitz ligament) + truncal vagotomy + sigmoid colon repair operations were performed. Drainage tubes were placed near the gastrojejunostomy and the pouch of Douglas. No complications due to the operation were observed after surgery. Drains were withdrawn on day 3, and the patient was discharged on day 5. At 1 month of follow-up, the patient's complaints decreased and he started to gain weight. At 6 months of follow-up, it was determined that there was no active complaint, that the laboratory values were normal, and that he received about 8 kilograms. The patient was granted permission to share medical information.

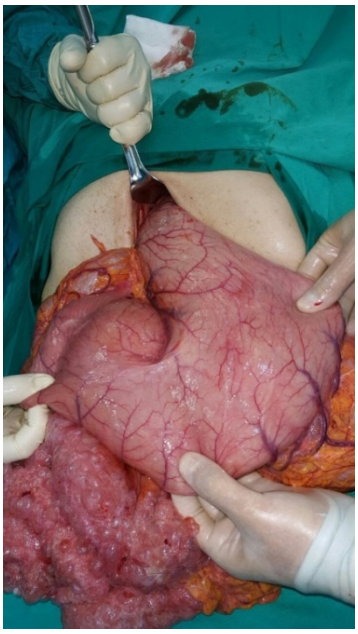

Figure 1. The stomach was found to be extremely dilated.

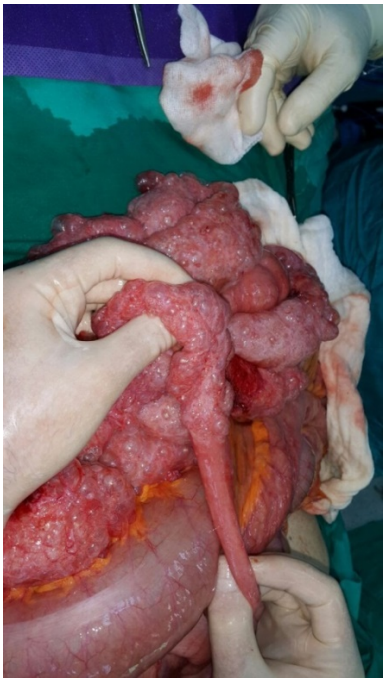

Figure 2. On the intestines cystic formations limited to the serosa were observed, affecting a 120 cm length of small intestine starting 1 meter after Treitz ligament.

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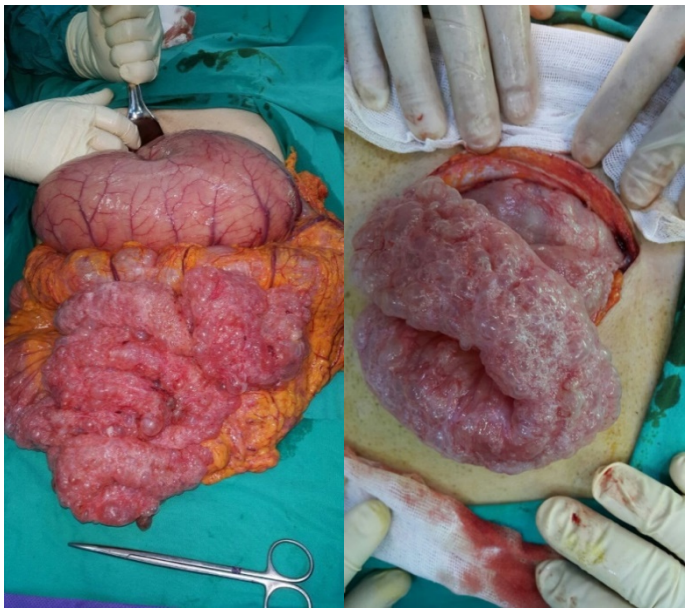

Figure 3-4. Exploration views of the operation.

DISCUSSION

Pneumatosis cystoides intestinalis is a rare pathology of unknown etiology that is characterized by numerous gas-filled cysts in the subserosa and submucosa layers of the intestines[1]. Overall prevalence of PCI is the same in men and women, and most cases occur in the 6th decade of life[10]. Our index case was a 62-year-old man. While jejunum and ileum are the most commonly involved areas, the colon is affected in 6% of cases[1]. Our patient had jejunoileal lesions. Many gastrointestinal system diseases such as appendicitis, Crohn's disease, pyloric stenosis, ulcerative colitis, diverticulosis, necrotizing enterocolitis, gastroduodenal ulcers and sigmoid volvulus may be associated with PCI. Cases of PCI secondary to surgical or endoscopic trauma were also reported[8].

The form of PCI not associated with any intestinal or systemic disease is called 'primary' (15%), and the form associated with a disease is termed 'secondary' (85%) [11,12]. Our patient had pyloric stenosis and underwent endoscopic balloon dilation three times, the last of which was performed three months ago. For this reason, our case was regarded as secondary PCI.

PCI patients are usually asymptomatic, but sometimes they present with non-specific complaints like abdominal pain, distension, diarrhea, constipation, weight loss and tenesmus[13]. Symptomatic patients are usually diagnosed while being evaluated for underlying diseases. In some patients, life-threatening clinical conditions such as NEC, septic shock, abdominal distension, partial obstruction, volvulus and intussusception may also be seen. Physical examination is often normal[5,12,14].

Non-specific symptoms such as diarrhea, constipation, rectal bleeding, tenesmus, weight loss and abdominal pain may be seen in PCI[9]. Complications such as volvulus, intestinal obstruction, tension pneumoperitoneum, hemorrhage, intussusception and intestinal perforation can be seen in 3% of patients[15,16]. This

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Geliş tarihi/Received: 25.04.2017 Kabul tarihi/Accepted: 30.07.2017

patient had weight loss and upper GI tract hemorrhage. Direct abdominal x-ray and abdominal CT are valuable imaging modalities for PCI[8]. However, radiological diagnosis can be very difficult if the disease takes a mild course and there is no pneumoperitoneum. This case of PCI was incidentally discovered during surgery. Since half of patients with PCI have spontaneous remissions, and because gas cysts are known to recur after surgery; no specific treatment is used in asymptomatic cases[1,8,10]. If symptomatic patients do not have signs of perforation, peritonitis or sepsis, non-operative oxygen therapy is first applied[8]. In our patient, we did not intervene surgically because there were no symptoms associated with PCI. Pyloric stenosis was thought to be the likely etiology of PCI in this case, while endoscopic trauma was also considered possible, but less likely. When free air in the abdomen is shown radiologically, the differential diagnosis should include PCI, in addition to intestinal perforation and bacterial infections that can produce gas.

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