Radiology Case Reports 15 (2020) 2459–2463

Available online at www.sciencedirect.com

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Case Report

Pneumatosis intestinalis with pneumoperitoneum: Not always a surgical emergency ☆,☆☆,★,★★

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ARTICLE INFO

Article history:
Received 15 June 2020
Revised 10 September 2020
Accepted 11 September 2020

Keywords:
Pneumatosis intestinalis
Pneumoperitoneum
Benign pneumatosis
Surgical emergency

ABSTRACT

Pneumatosis intestinalis (PI) and pneumoperitoneum are commonly recognized as severe signs of gastrointestinal diseases that require emergency surgery. However, these symptoms can also be caused by benign conditions. We describe 4 cases of benign PI and pneumoperitoneum that were detected in different clinical situations (accidental discovery in bilan of aortic dissection (case #1), bilateral pulmonary embolism (case #2), overflow diarrhea due to fecal impaction (case #3), and in follow-up postbiliary digestive anastomosis surgery (case #4), which were addressed with exploratory surgery (case #1) or conservative treatment (the remaining cases), with favorable outcomes. Because PI and pneumoperitoneum can be associated with both life-threatening causes and benign conditions, treatment decisions should be based on the correspondence between clinical and paraclinical features, rather than imaging alone.

☆ Funding: This study was not supported by any funding.
☆☆ Declaration of Competing Interest: There are no conflicts of interest to declare.
★ Ethical approval: Owing to design of case series, institutional review board approval was waived.
★★ Declaration of patient consent: Informed consent of patients was obtained.
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https://doi.org/10.1016/j.radcr.2020.09.034

Introduction

Pneumatosis intestinalis (PI) describes abnormal intramural gas of the digestive tract. Pneumoperitoneum refers to the presence of gas in the peritoneal cavity, out of the digestive tract. The presentation of these 2 symptoms together generally suggests the occurrence of life-threatening conditions, such as gastrointestinal perforation, ischemic bowel, and bowel necrosis [1–3]. However, some benign conditions can also cause this phenomenon, such as respiratory diseases, systemic diseases, immunodeficiency, several medications, postoperative, post-transplantation organs, and iatrogenesis [4–7].

We describe 4 cases of PI with pneumoperitoneum that were associated with 4 distinct causes. One case underwent exploratory surgery to rule out surgical causes, whereas the remaining 3 cases were managed conservatively, with effective outcomes.

Case report

Case 1

A 58-year-old man was hospitalized with acute chest pains, vomiting, and nausea. His medical history included: hypertension, treated by propranolol and perindopril; dyslipidemia; chronic gout, treated by Allopurinol; and chronic obstructive pulmonary disease, treated by Spiriva 18 and Novopulmon. On examination, he appeared uncomfortable and diaphoretic, with a heart rate of 120 bpm, blood pressure of 167/74 mm Hg, and room-air peripheral capillary oxygen saturation (SpO_2) of 96%. His abdomen was mildly distended, without abdominal guarding or rebound tenderness.

A contrast-enhanced thoraco-abdominopelvic computed tomography (CT) was performed showing an aortic dissection originating from the ascending aorta, with involved dissection of the brachiocephalic artery, extending to the right common iliac artery (Stanford type A). The superior mesenteric artery, celiac trunk, and right renal artery originated from the true lumen. The inferior mesenteric artery and the left renal artery originated from the false lumen, with effective enhancement. Pneumatosis of the left colonic angle and pneumoperitoneum were observed (Figs. 1a and b). The evaluation of the colonic wall enhancement was difficult, due to diffuse PI. Neither portomesenteric venous gas nor ascites were identified.

The patient underwent exploratory laparotomy, and no surgical lesions causing gas in the intestinal wall or peritoneum were detected. Afterward, he underwent supracoronary ascending aortic replacement. The postoperative state was stable, and metronidazole was given for 7 days. The patient was discharged, without major complication, peritonitis, or gastrointestinal abnormalities.

Postoperative evaluation CTs at 1 and 3 months showed the stable state of the aortic dissection, and no air in the wall of the colon or peritoneal gas was detected (Figs. 1c and d). However, CT at 6 months detected a diffuse PI along the entire descending and sigmoid colon, which spread to the rectum, with no fat stranding, nondilated colon, and no fluid or pneumoperitoneum (Figs. 1e and f). Because he was asymptomatic,

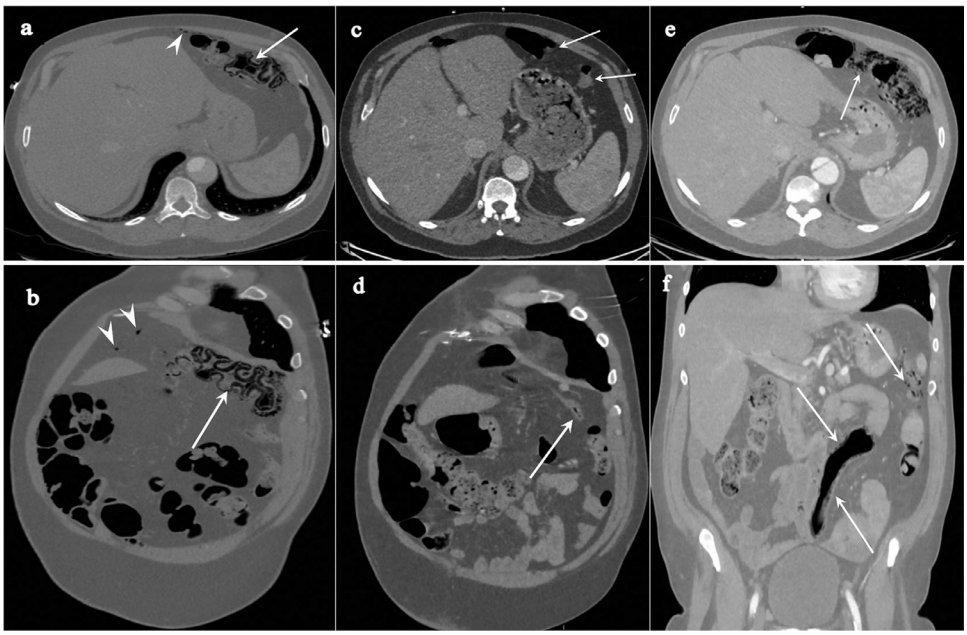

Fig. 1 – Case 1. Contrast-enhanced abdominal CT. (a and b) Axial and coronal oblique, at the time of admission, showing diffuse PI along left colonic angle (arrow) and adjacent pneumoperitoneum (arrowhead). (c and d) Axial and coronal oblique, 1 month after surgery: Neither PI (short arrow) nor pneumoperitoneum was observed. (e and f) Axial and coronal oblique, 6 months after surgery: diffuse PI along the descending and sigmoid colon (arrow); no pneumoperitoneum.

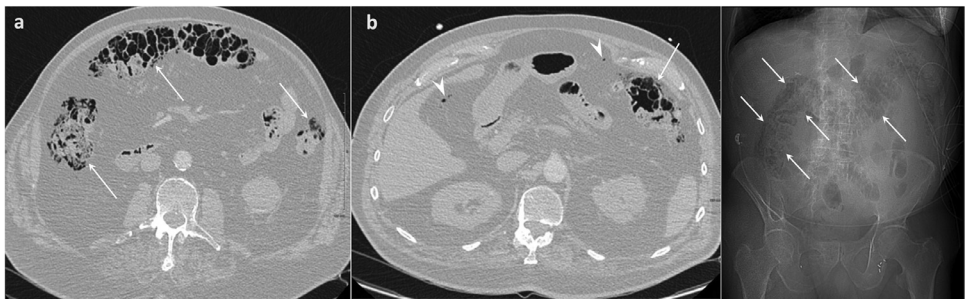

Fig. 2 – Case 2. Contrast-enhanced abdominal CT, lung window: a diffuse PI of the left colonic angle (arrow) and pneumoperitoneum (arrowhead) (a and b). Plain film abdominal, showing extensive PI along the entire ascending, transverse, and descending colon (c).

a recurrence benign PI was diagnosed, but no treatment required.

Case 2

A 78-year-old man was hospitalized due to the sudden onset of continuous severe dyspnea during doing exercise, which did not reduce at rest, accompanied by constrictive chest pain. On admission, he was uncomfortable, diaphoretic, and frightened. His blood pressure was 150/88 mm Hg, his heart rate was 93 bpm, without murmurs, and he was tachypneic, with an air-room SpO_2 of 94% (3 L/min). His chest was clear on examination and the abdominal examination was unremarkable.

Blood tests showed D-dimer >4000 ng/mL, troponin levels of 41 ng/L, N-terminal pro-brain natriuretic peptide (NT-proBNP) levels of 2870 pg/mL, and serum lactate levels of 1.5 mm/L. Contrast-enhanced CT detected a bilateral, massive, proximal pulmonary embolism, with an impact on the right ventricle. No pleuro-pericardial effusion was detected. However, PI of the colon, predominant on the right side, with some pneumoperitoneum bubbles were detected, without atypical bowel wall enhancement (Fig. 2). No fat stranding, ascites, or other abnormal signs were detected. Afterward, on complement tests, an acute deep venous thrombosis was detected.

The patient was treated for pulmonary embolism, and followed bowel sounds. He was discharged 10 days later, without adverse events.

Case 3

An 82-year-old woman was admitted to the hospital for abdominal pain, diarrhea lasting for 3 weeks, signs of dehydration, following antibiotic administration 4 weeks prior to admission. She presented with loose, watery stools, but no bloody stools. She also complained of lost appetite and had lost more than 10 kg during the previous month. Past medical history included breast cancer status post mastectomy and chemoradiation, and depression.

On examination, she was afebrile, with normal blood pressure, a pulse of 110 bpm, SpO_2 of 97%, and no sign of severe dehydration. On abdominal examination, her bowel sounds were active, with slight tenderness and some hard stool in the region of the left colon. No other abnormalities were identified. Her blood tests revealed a C-reactive protein level of

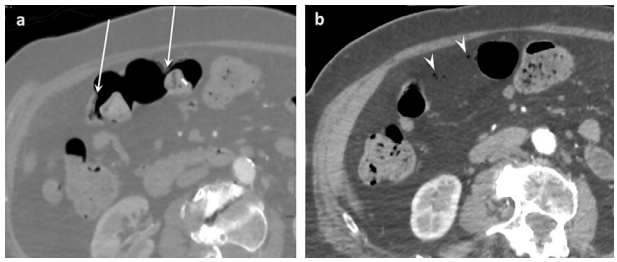

Fig. 3 – Case 3. Contrast-enhanced abdominal CT, showing linear PI along the Sigmoid colon (a) (arrow) and several bubbles of pneumoperitoneum (b) (arrowhead).

23 mg/L, no anemia, mild electrolyte disorders (sodium level of 134 mmol/L, potassium of 3.4 mmol/L, chloride level of 98 mmol/L), a normal level of serum lactate, and normal renal function. The stool culture was negative, even for *Clostridium difficile*. Contrast-enhanced abdominal CT showed a stercoral stasis, with large rectal fecal impaction and mild adjacent fat stranding, without signs of intestinal obstruction syndrome. Colonic pneumatosis of the sigmoid loop, with a few bubbles of pneumoperitoneum, were observed, without infiltration of adjacent fat (Fig. 3). No portomesenteric venous gas, no ascites, and no abnormality of the mesenteric vessels were detected.

The diagnosis was overflow diarrhea due to fecal impaction, with benign PI and pneumoperitoneum. The patient was treated with fluids and electrolyte management, laxative treatment (MACROGOL, 5.9 g twice a day), and supplementations for folate deficiency and vitamin D. Symptoms quickly improved, with several episodes of loose stools during the first day, which then returned to normal. Abdominal CT was not indicated after treatment.

Case 4

A 67-year-old woman was followed up in our hospital for a biliary neuroendocrine tumor, which was resected with a Roux-en-Y anastomosis 7 years before. A few days before admission, she had vomited several times and noticed recent weight loss. However, the patient was in very good general condition, with no other symptom. Physical examination was un-

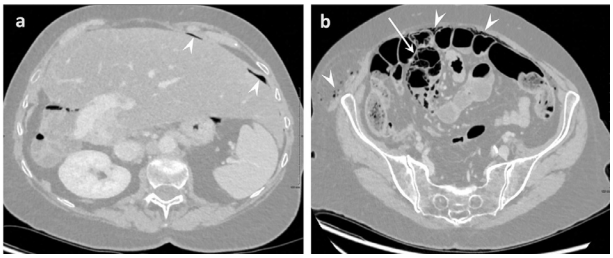

Fig. 4 – Case 4. Contrast-enhanced abdominal CT (a and b) demonstrated a diffuse PI of the small bowel (arrow) and scatted pneumoperitoneum (arrowhead).

remarkable. A follow-up thoraco-abdominopelvic CT was performed, showing stable disease according to RECIST 1.1; however, diffuse PI of the small intestine, especially in loops near the anastomosis, and scatted pneumoperitoneum were detected (Fig. 4). No signs of mesenteric infiltration or ascites were observed. Based on the symptomatology and the CT results, she was hospitalized to monitor clinical signs and take paraclinical tests (complete blood count, electrolyte test, renal and hepatic functional tests, blood gas test) which detected no abnormalities, no inflammatory syndrome, normal serum lactate level, and normal hepatic and renal function were preserved. No procedure was performed.

Given the very good clinical and biological evolution, she was allowed to return home the next day.

Discussion

PI is rare and often localized in the submucosa or subserosa. When combined with pneumoperitoneum, PI usually indicates a surgical emergency [2,3]. However, some nonemergency causes that can lead to PI have been described, such as respiratory diseases, systemic diseases, immunodeficiency, after organ transplants, and iatrogenesis [4–6].

The physiopathology of PI remains unclear. Four hypotheses regarding this process have been proposed: bowel necrosis, gas from the intraluminal gastrointestinal tract, gas-producing bacteria, and gas originating from alveoli [5–9]. In the first 2 cases, respiratory diseases are the most likely causes of PI and pneumoperitoneum. Some authors have suggested that high interalveolar pressure associated with lung diseases may result in the emanation of gas following alveolar rupture, which spreads intermittently to the mediastinum and then descends into the retroperitoneal cavity and mesenteric roots before reaching the intestinal wall or peritoneal cavity [10]. A study in patients with cystic fibrosis and PI showed that 95% of patients presented with pulmonary interstitial gas or pneumomediastinum [11]. However, this theory does not explain cases in which methane or hydrogen is found in parietal bowel bubbles, whereas these gases are neither present in the lungs nor created by human cells but analogous to gas in the intraluminal bowel [12]. Another theory suggests that patients with pulmonary diseases commonly present with increased intra-abdominal pressure, especially during coughing or exertion, which can introduce gas into the intestinal mucosa gap [13]. This mechanism may explain how our second patient develops PI and pneumoperitoneum. Long-term corticosteroid therapy may also be a risk factor for this phenomenon, due to the weakening of the intestinal mucosa barrier [8]. In our third case, the patient had overflow diarrhea following constipation. This condition may cause damage to the mucosa of the colon, allowing bacteria to invade and produce gas in the intestinal wall [9]. Mechanical causes, such as the disruption of the gastrointestinal tract mucosa caused by ulcers, trauma, intervention, or surgery, can allow gas from the intraluminal region to diffuse to the parietal digestive tract [14]. This mechanism would explain our fourth case. Pneumoperitoneum may be caused by the direct diffusion from interstitial tissue or the rupture of air bubbles subserosa [15].

Clinically, benign PI is often an asymptomatic, incidental finding during endoscopy or imaging examinations [4,16]. Sometimes, PI presents mild symptoms, including abdominal discomfort, diarrhea, and hematochezia [1]. Severe clinical symptoms are generally observed in surgical emergencies such as ischemia, bowel necrosis, intestinal volvulus, and peritonitis [2,4,8]. However, in some clinical circumstances, the distinction between a surgical and nonsurgical situation may be difficult, due to overlap with other pathologies. In our first case, the patient was hospitalized, due to the presentation of acute ischemia risk factors. Although the clinical symptoms were unclear, laparoscopic surgery was performed to exclude a colonic infarction. The remaining 3 cases only presented with mild clinical symptoms or were asymptomatic, and surgery was not indicated.

CT imaging is a common examination modality used to detect PI and pneumoperitoneum, as well as other signs of digestive injury [9,16]. Some authors have reported several imaging signs that can differentiate between benign and surgical groups. The absence of bowel enhancement is a specific finding associated with ischemia that occurs during the late stages [2]. Bowel wall-thickening, mesenteric fat-stranding, and ascites are prominent findings that indicate a surgical situation, but these symptoms can also be observed in nonemergency patients [1,4,9]. Hawn et al. revealed that a serum lactate level >2 mmol/L suggested a risk of mortality greater than 80% [17] and may represent a good marker for the distinction between benign and life-threatening situations.

Corticosteroid therapy, antibiotics, and oxygen therapy may be useful to reduce the symptoms that have been revealed in some reports [6,13,18]. Oxygen therapy should be maintained for at least 48 hours after the disappearance of PI, to avoid recurrence [13]. For asymptomatic patients, conservative treatment is proposed [6,19]. Surgery is only indicated in cases of complications, such as perforation, volvulus, and ischemia bowel [20].

Conclusions

PI and pneumoperitoneum can be associated not only with life-threatening situations but also with benign situations. Compatibility between clinical and paraclinical elements are

necessary to provide appropriate treatment. In uncertain cases, laparoscopy is a simple procedure that can be performed to rule out surgical causes.

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