

A Case of Benign Pneumatosis Cystoides Intestinalis (PCI): what the Radiologist Should Know about PCI and life-Threatening Pneumatosis Intestinalis (PI)

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ABSTRACT

Pneumatosis cystoides intestinalis (PCI) is a rare entity characterized by the presence of gas within the intestinal wall; it may be classified as primary/idiopathic or secondary.

PCI usually remains asymptomatic and often represents an imaging incidental finding.

The presence of symptoms mostly depends on the site of bowel involvement, mass effect or related complications. Imaging still plays a key-role not only in establishing the correct diagnosis but also in assessing the presence of comorbidities or complications.

Ultrasound (US) is mostly used in the pediatric population while in the adult population X-rays may be the first approach. Computed tomography (CT) is always the preferred study for diagnosis, differential diagnosis and follow-up.

It allows accurate visualization of intramural gas in cystic or linear configuration and helps avoid unnecessary surgical intervention by identifying associated high-risk features.

The aim of this case report is to show a rare benign situation of PCI in a young man who presented to our emergency room (ER) after a blunt trauma.

Keywords

Pneumatosis cystoides intestinalis (PCI), Pneumatosis intestinalis (PI).

Introduction

Pneumatosis intestinalis (PI) is an imaging finding characterized by the presence of gas within the wall of the intestine.

This finding is associated with numerous conditions, ranging from benign to life-threatening situations [1]; mesenteric ischemia and bowel obstruction represent the most common life-threatening causes of PI [2].

Other life-threatening causes of PI include cecal ileus, toxic megacolon, collagen vascular diseases, necrotizing enterocolitis, neutropenic colitis, pneumoperitoneum, volvulus and sepsis [1-4].

The overall incidence of PI in the general population has been reported to be 0,03% [1].

Two patterns of PI have been described [2]:

- A linear pattern in which the gas has a curvilinear and circumferential form within the bowel wall [2]; the presence of this pattern with additional specific findings such as abnormal mucosal enhancement, distended bowel, arterial or

venous occlusion, ascites, pneumoperitoneum and portal or porto-mesenteric venous gas, increases the risk of PI due to a life-threatening cause [2].

- A bubble-like or cystoid pattern characterized by separate bubbles of gas and a cystic appearance within the intestinal wall layers. PCI is considered a rare entity, commonly affecting the small intestine and colon but can occur from the esophagus to the rectum [4].

It usually affects adults in the fourth to sixth decades of life, with equal distribution between males and females [3]; it may be either primary/idiopathic or secondary.

The primary or idiopathic type represents 15% of cases and is considered a self-limited or chronic benign entity [3].

The secondary type represents 85% of cases and is associated with several factors, non gastrointestinal causes as well, such as surgery, pharmacotherapy, chemotherapy, autoimmune diseases, hematological malignancies, inflammatory diseases, acquired immune deficiency syndrome, obstructive airways disease or pulmonary illness such as cystic fibrosis and asthma [1-4].

While the exact pathophysiology of PCI remains unclear, it is believed to be related to one or more of the following: Mucosal injury, gas producing bacteria and conditions with reduced mucosal integrity and altered intestinal flora [4].

Furthermore, the Macklin effect could represent a crucial secondary, extra-gastrointestinal mechanism of PCI. This phenomenon involves alveolar rupture leading to the centripetal dissection of air along the bronchovascular sheaths into the mediastinum (pneumomediastinum), from where it can track retroperitoneally into the mesentery and seamlessly infiltrate the subserosal or submucosal layers of the bowel wall. This barogenic mechanism is frequently triggered by a sudden increase in intra-alveolar pressure during blunt thoracic trauma, or as a consequence of barotrauma in COVID-19 patients undergoing invasive or non-invasive mechanical ventilation due to virus-induced lung parenchyma frailty [5].

PCI is usually asymptomatic, frequently representing incidental clinical or radiological finding.

If present, clinical findings of PCI are often non-specific and abdominal pain, distension and diarrhea may be the most common symptoms [4]. Presence of symptoms depends on the site of involvement, mass effect or related complications.

In fact, most symptomatic PCI depend on secondary intestinal obstruction because of its mass effect in large size cysts or pneumoperitoneum due to rupture of the cysts, causing in these cases symptoms like abdominal pain, swelling and general discomfort; however, complications are not so common.

For this reason, imaging plays a pivotal role not only in establishing

the diagnosis but also in assessing disease extension and ruling out complications.

Ultrasound (US) is mostly used in the pediatric population while in the adult population X-rays may be the first approach.

Computed tomography (CT) is always the preferred study for diagnosis, differential diagnosis, follow-up, helpful in avoiding unnecessary surgery treatment, best demonstrating multiple cysts or linear gas within the serosa and sub-serosa of the intestinal wall and any complications occurred.

CT is also effective in evaluating the correct management, by assessing the intestinal level involved, changes in the size, number and shape of them, ranging from purely cystic lesions to a mass-like aspect.

In PCI cysts are usually multiple but unilocular.

The aim of this case report is to show a rare situation of PCI in a young man who presented to the emergency room (ER) after a blunt trauma.

Case Presentation and Clinical History

A 32-year-old man presented to the ER after a blunt trauma mainly involving the head, without abdominal or chest pain or visible bruises.

He was alert and cooperative at triage, he had a normal acid-base balance on venous blood-gas analysis and no nausea, diarrhea nor fever; laboratory tests were unremarkable, with normal white blood cell count, c-reactive protein and serum lactate.

No relevant clinical findings were found related to the trauma or to his previous clinical history as well.

Imaging Findings

After the triage, a total-body CT scan was performed. Abdominal CT revealed an organized cluster of cystoid formations with air content in the layers of colic intestinal loops at the level of the junction between the transverse colon and its splenic flexure, for a longitudinal extension of at least 10 cm (Figures 1a, b, c, d).

Isolated free air bubbles within the surrounding adipose tissue were found and attributed to the rupture of serosal and sub-serosal cysts in the transverse colon wall (Figure 2).

No free fluid, bowel wall thickening or signs of obstruction were detected.

Based on imaging findings and clinical stability, a diagnosis of benign PCI was established.

Head and chest CT scans were negative.

The patient was then carefully monitored and according to the

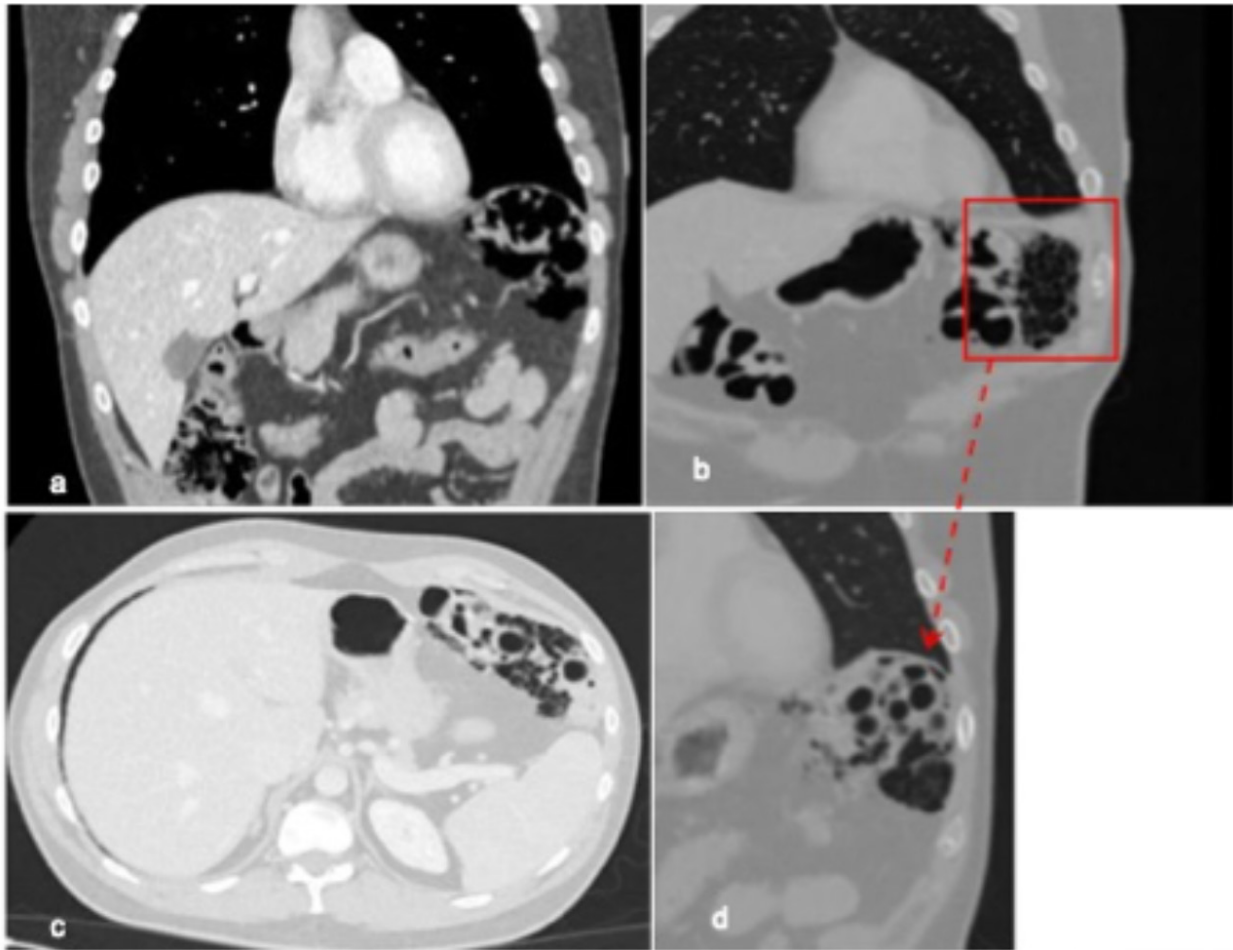


Figure 1a,b,c,d: Abdominal CT images show an organized cluster of cystoid, air-filled formations within the wall of the colic intestinal loop at the junction between the transverse colon and its splenic flexure, extending longitudinally for at least 10 cm.

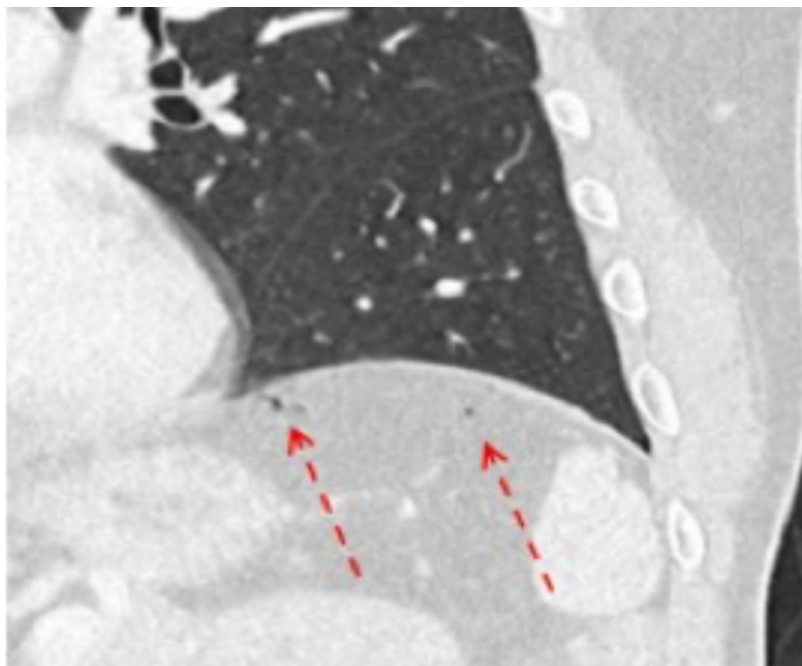


Figure 2: Isolated free air bubbles are visible within the surrounding adipose tissue, attributed to the rupture of serosal and sub-serosal cysts in the transverse colon wall.

diagnosis of PCI, no relevant clinical changes happened in the following 24 hours of observation.

In this case, a non-operative approach was then considered. Follow-up over time was anyway suggested to him if he remained clinically well.

Discussion

The diagnosis of PCI is based on clinical findings, laboratory values and imaging features where CT scan represents the most sensitive radiological exam.

PCI is a gastrointestinal disease but it mostly affects the colon (80%) followed by the small bowel.

Diagnosis has however largely been based on radiological investigation, even if endoscopy plays an important role for the direct visualization of sub-mucosal cysts which present as polypoid structures [4].

In PCI, CT identifies the intramural gas and any signs of spontaneous perforation including pneumoperitoneum, pneumoretroperitoneum or mesenteric gas, best demonstrated in lung window [4] even only in a basal CT evaluation; over-infection or intestinal obstruction due to cysts mass-effect may also occur. Management of PCI relies on differentiating between benign and life-threatening PI.

It means differentiating between benign and life-threatening PI causes, first of all intestinal ischemia, particularly in a setting of peritonitis, hemodynamic instability or leukocytosis [4].

Laboratory values during intestinal ischemia may reveal acidosis (pH <7.3), hyperamylasemia (>200 IU/L), reduced bicarbonate level (<20 mmol/L) and elevated serum lactic acid (>2 mmol/L).

Free fluid, reduced bowel wall enhancement, bowel wall edema and portal venous gas are more frequently seen in life-threatening PI and strongly suggest the need for an operative approach.

According to it, all of these features were absent in the presented case.

In fact, asymptomatic patients require no intervention and may be managed conservatively with supplemental oxygen including hyperbaric oxygen, facilitating the diffusion of gas from the cysts into the blood stream [4]; antibiotics may also be added. Sometimes, endoscopy can be used therapeutically to facilitate aspiration or excision of cysts [4].

The incidence of complications of PCI such as perforation or obstruction is reported as 16,3% [4].

Surgical treatment should be reserved for patients who remain symptomatic despite medical therapy or who develop PCI-related complications such as peritonitis, perforation, sepsis, necrosis or recurrent bowel obstruction [2].

Conclusion

PCI predominantly affects the colon, followed by the small bowel, but it can potentially affect almost any site of the gastrointestinal tract, with different extension, imaging features and clinical discomfort.

Commonly CT demonstrates characteristic imaging findings but sometimes it can create diagnostic challenges mostly mimicking life-threatening conditions of PI in which a surgery approach is mandatory.

As symptoms are non-specific or absent altogether, imaging still plays a crucial role.

Owing to its rarity and absent or non-specific symptoms, PCI can be easily misdiagnosed.

This case highlights the importance of recognizing PCI quickly, as benign as it is an uncommon condition, and the role of non-operative management in order to reduce unnecessary surgery and potential morbidity related to it.

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