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Case Report

Conservative Treatment in Pneumatosis Cystoides Intestinalis: A Case Report and a Brief Literature Review

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Abstract

Introduction: Pneumatosis Cystoides Intestinalis (PCI) is a rare condition in which submucosal or subserosal gas cysts are found in the wall of the small or large bowel. Many different causes of pneumatosis cystoides intestinalis have been proposed, including mechanical and bacterial causes, but still today the real pathogenesis is unclear.

Case presentation: We described a case of a patient with pneumatosis cystoides intestinalis treated with a conservative approach.

Discussion: The pneumatosis cystoides intestinalis is a rare disease and suspicion of this disease process should be based on imaging and clinical finding. The therapy can be conservative or surgical in restricted situations.

Conclusion: For patients with PCI, a conservative approach is the treatment of choice, while surgery is mandatory only for complicated disease.

Keywords: Pneumatosis cystoides intestinalis; Pneumatosis intestinalis; Abdominal pain; Portal system; Surgical treatment; Conservative treatment.

Abbreviations: PCI: Pneumatosis Cystoides Intestinalis; PM: Pacemaker; MDCT: Multidetector Computed Tomography; CT: Computerized Tomography; MRI: Magnetic Resonance Imaging; LDH: Lactate Dehydrogenase.

Introduction

Pneumatosis intestinalis is the presence of gas-filled cysts in submucosa or subserosa of small or large bowel wall and is divided into two categories: life-threatening pneumatosis intestinalis and benign pneumatosis intestinalis. Distinguishing between pneumatosis cystoides intestinalis and life-threatening pneumatosis intestinalis may be challenging, although computed tomography scan allows the detection of additional findings that may suggest an underlying, potentially cause of pneumatosis intestinalis. To correctly manage the patients affected by this disease is important to differentiate the two types of pneumatosis and investigate the correct pathogenesis that nowadays is still unclear altought some causes have been theorized.

According to literature, approximately 85% of cases are thought to be secondary to coexisting mechanical or bacterial disorders of the gastrointestinal tract or the respiratory system [1].

The mechanical theory, which is the most accepted explanation, suggests that gas under pressure is forced into the bowel wall through a mucosal defect associated with trauma, surgery, endoscopy and bowel obstruction.

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Second, there is the bacterial theory. In animal experiments, introduction of bacteria into the bowel wall by injection has been shown to cause PCI. The pulmonary theory has been criticized as the assumption that gas travels from ruptured alveoli through the mediastinum into retroperitoneal space and finds its way along perivascular spaces through the mesentery into the bowel wall could not be proven convincingly [2].

The presenting clinical findings may be very heterogeneous and symptoms of pneumatosis cystoides intestinalis, depending on the location of the gas filled cysts, may include diarrhea, constipation, rectal bleeding, mucorrhoea, abdominal discomfort, abdominal pain, urgency, malabsorption, weight loss and excessive flatus. Depending on the location of the gas filled cysts the range of symptoms in each patient may vary enormously [3]. Intestinal pneumatosis may lead to various complications. The patients with pneumatosis cystoides intestinalis are usually treated conservatively; the surgical treatment is reserved for complications [4].

In this case report we evaluate the safety and the risk when a conservative approach is applied in patients with PCI in accordance to the guidelines mentioned in "International Committee of Medical Journal Editors" [5].

Case presentation

The case we are describing it is about a 81-year-old woman with abdominal pain, nausea, vomiting, abdominal distention and discomfort for 4 days. She did not develop fevers.

Her blood chemistry tests showed no alterations in the inflammation indices, but only a slight increase in LDH and lactates.

She was admitted from our emergency room to our radiology department for further evaluation of her symptom.

The medical history of the patient revealed arterial hypertension, mitral valve replacement for stenosis with mechanical valve, atrial fibrillation, a PM, chronic renal failure, hypothyroidism; the patient did not report a history of gastrointestinal desease. The abdomen CT showed small share of air in the peripheral intrahepatic portal system, with arboriform appearance (Figure 1).

Slight fluid distension of ileal loops located in the meso-hypogastric, some of which with thickened walls with mucosal hyperemia and others with probable wall pneumatosis and associated air in the context of some respective mesenteric vascular structures" (Figure 2).

The above-described findings were first of all due to acute intestinal distress.

Upon admission to the hospital, the patient's abdomen was treatable and her abdominal pain had subsided. The biochemical parameters did not show substantial variations compared to those performed in the emergency room, so it was decided to postpone an emergency video laparoscopy and to continue the clinical observation. She was managed with bowel rest, nasogastric tube decompression, hydration and broad-spectrum antibiotics.

The day after, the biochemical parameters improved and the abdomen CT with contrast agent reported the following "no longer appreciable the share of air in the peripheral intrahepatic portal system. Slight and fluid distension of the ileal loops is confirmed with probable reduced wall pneumatosis compared to the control" (Figure 3).

The finding of pneumatosis intestinalis resolved over the ensuing 3 days.

Her diet was slowly advanced after 4 days of fasting and she was discharged home in stable condition without further surgical intervention or recurrence of the pneumatosis.

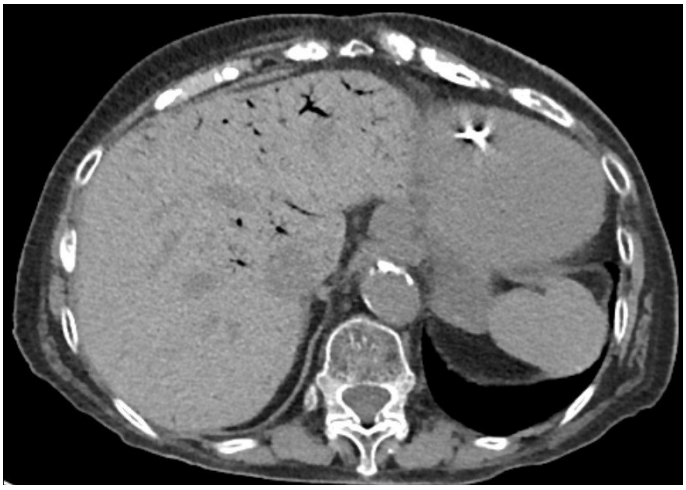

Figure 1: Clinical image.

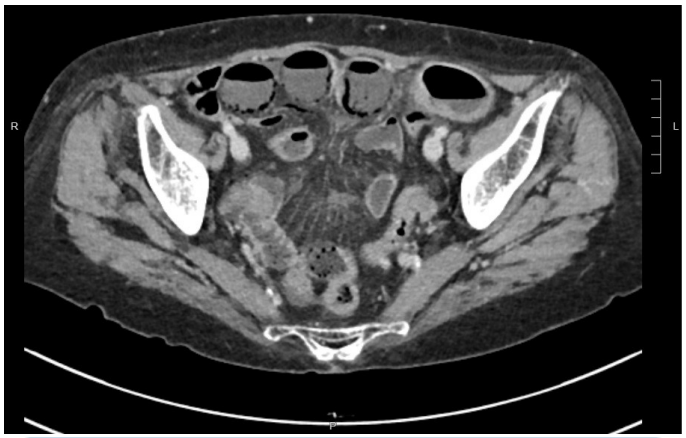

Figure 2: Clinical image.

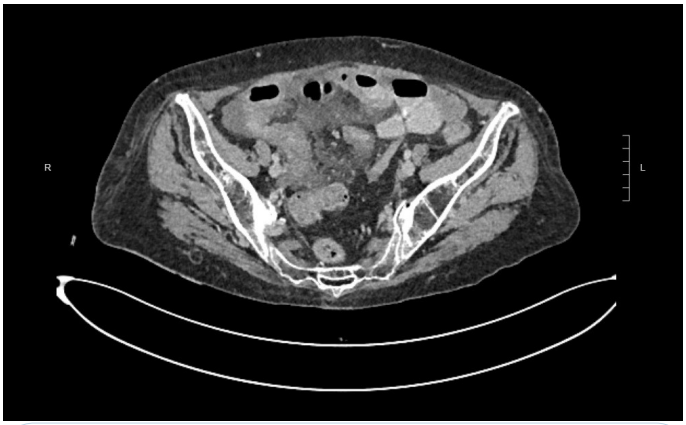

Figure 3: Clinical image.

Discussion

Pneumatosis intestinalis was defined by Lerner and Gazin in 1946 as the presence of gas in an abnormal site of the body [6]. PCI is a rare condition characterized by multilocular gas-filled cysts localized in the submucosa and subserosa of the gastrointestinal tract. The data present in the literature do not currently allow us to ascertain the real incidence. Symptoms, if any, are abdominal pain, diarrhea, constipation, rectal bleeding, tenesmus or weight loss and severe complications, including volvulus, intestinal obstruction, tension pneumoperitoneum, bleeding, intussusception, and intestinal perforation, may be seen. Pneumoperitoneum and pneumoretroperitoneum can be rare complications due to rupture of the cysts. In our case, free intraperitoneal air was secondary to a mechanical cause maybe because of an intestinal occlusion by an intestinal volvulus subsequently resolved spontaneously.

Radiological tools as plain radiographs, ultrasonography, barium series, MDCT, MDCT colonoscopy and MRI are important for diagnosing PCI. In particular x-ray is of great importance because it is readily available in every emergency room but only MDCT give us higher quality and accuracy because of the advancement of multidetector technology. Cysts usually appear as radiolucent shadows, similar to a bunch of grapes, close to the intestinal lumen on radiographs on the contrary MDCT show spatial resolution and is the most useful modality for diagnosing PCI and other intra-abdominal pathologies. To confirm a PCI diagnosis is useful surgical exploration if the physical examination and imaging findings are suspicious [6].

Conclusion

The clinical condition of the patient, not only the finding of pneumatosis intestinalis, should drive management in these cases [3,7].

In conclusion, as we illustrated by this case report, the correct diagnosis and management of PCI is based on the results of clinical assessment and imaging techniques. PCI also should be kept in mind as a rare cause of pneumoperitoneum. Concordantly, it is very important for the radiologist to recognize the abnormal findings on the MDCT or MRI studies for differentiating between medical and surgical causes of PCI. Conservative approaches, including nasogastric decompression, intestinal rest, antibiotic therapy and oxygen are recommended for patients with positive examination findings and normal biochemical parameters who are confirmed radiologically to have no intestinal ischemia or perforation. An urgent laparotomy is necessary in cases of intestinal ischemia, obstruction, intestinal bleeding, or peritonitis related to high mortality rate due to pneumatosis cystoides intestinalis. As a result, many authorities advocate an aggressive surgical approach in those patients so definitive surgery should be performed during laparotomy if necrosis, perforation or marked ischemia is observed in the intestine, but no in our patient [2].

In this case report we show the opportunity of the “no treatment”. The procedure is feasible and the choice of the strategy to be employed should be individualized based on diagnosis, patient characteristics, availability of resources and experience of surgical team.

Declarations

Consent for publication: Written informed consent was obtained from the patient for publication of this case report.

Ethics approval and consent to participate: Ethical approval was not applicable.

Conflict of interest: The authors declare that they have no conflict of interest.

Financial disclosure: The authors declared that this study has received no financial support.

Authors’ contributions: Bonventre Giulia provided the preparation of manuscript, the conceptualization and planning of the case report. D’Avolio Michele contributed to the diagnosis and management of the case. Mingoia Giovanni, Dominici Domenico Marco, Nicosia Giuseppe , Sferrazza Sonia , Raspanti Cristina , Ferrara Gabriella , Palma Antonio , Maltese Stefania contributed to the data collection. Di Gregorio Riccardo contributed to the figures selection. Every authors read and approved the final manuscript.

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