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Case Report

PNEUMATOSIS CYSTOIDES INTESTINALIS DURING THE TREATMENT WITH PACLITAXEL FOR METASTATIC OVARIAN CANCER

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ABSTRACT

Gas within the bowel wall can be an incidental finding in Computed Tomography (CT) exams, with increased frequency in oncological patients. Pneumatosis cystoides intestinalis (PCI) is an unusual subtype of this condition which mainly affects the colon, and which is characterized by circular air collections in the bowel wall. We report a case of a 74-year-old woman treated with Paclitaxel for metastatic ovarian cancer, in which a restaging CT scan showed PCI involving the right colon with microperforation. Subsequent CT performed after changing the therapeutic regimen showed a reduction in these findings. In oncological patients, PCI can be an incidental finding and can be safely managed conservatively.

1. Introduction

Pneumatosis intestinalis (PI), defined as the presence of gas within the wall of the gastrointestinal tract, represents an imaging sign rather than a specific diagnosis (1). This condition is associated with a diverse spectrum of causes, ranging from benign to life-threatening (2). We have recently witnessed an increasing detection rate, probably due to the increased utilization of diagnostic cross-sectional imaging, mainly Computed Tomography (CT), in which this imaging phenomenon is detected.

Moreover, an increased incidence of PI may also result from the diffusion of relatively new medications, surgical procedures and medical treatments (2, 3).

Pneumatosis cystoides intestinalis (PCI) is an uncommon and almost always benign subset of PI, characterized by circular collections of gas in the gastrointestinal wall and the mesentery, most commonly affecting the colon (2, 4).

2. Case presentation

A 74 year-old woman, a housewife, receiving chemotherapy for a metastatic high-grade ovarian carcinoma since March 2017, presented at our Radiology department to undergo a restaging whole-body CT scan in June 2019. She had a two-decade history of cirrhosis secondary to chronic hepatitis complicated with portal hypertension. The patient's chemotherapy regimen at that time included Paclitaxel with a dose reduction of 20% due to comorbidities (86.4 mg/m^2), administered from June 2018 after progression from the previous treatment of carboplatin in monochemotherapy.

Physical examination and anamnesis, performed before the scheduled tomography, revealed no significant symptoms or signs, and in particular there was neither abdominal pain nor distension, and bowel movements were regular.

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Contrast-enhanced CT scan demonstrated multiple partially calcified peritoneal implants, consistent with peritoneal carcinomatosis, and a complex pelvic mass arising from the right ovary (Figure 1); the disease appeared stable compared to a previous CT performed 6 months before according to RECIST 1.1 criteria (5).

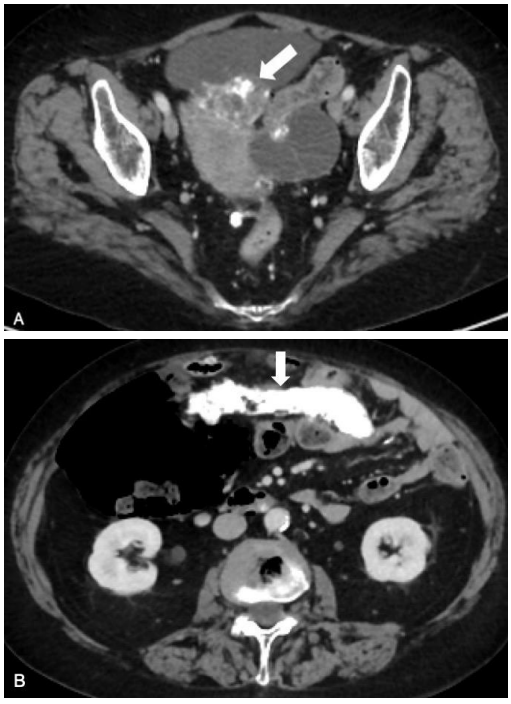

Figure 1. Axial contrast enhanced CT images in portal venous phase from helical CT scan in June 2019. Pelvic CT scan (A) showed a right ovarian mass (arrow) consisting in both solid and cystic components, whereas in upper slices (B) a partially calcified peritoneal implant (arrow) along the greater omentum was present, sign of large peritoneal carcinomatosis.

The scan was acquired with 64 detectors GE Lightspeed VCT scanner (General Electric Medical Systems, Milwaukee, WI) after a peripheral endovenous administration of contrast medium (95mL of Omnipaque 350mgI/mL) using automatic bolus tracking to obtain an arterial phase of the whole abdomen followed by a portal phase scan including neck, thorax, abdomen and pelvis.

Furthermore, CT revealed intestinal wall pneumatosis involving the right colon, from the cecum to the hepatic flexure, and a few retroperitoneal small free air bubbles (Figure 2).

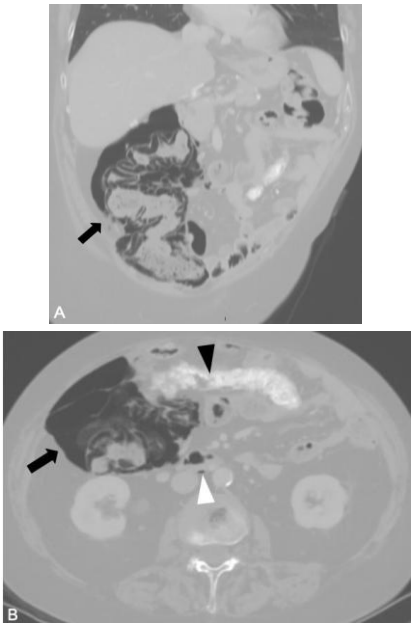

Figure 2. Coronal oblique multiplanar reconstruction and axial CT images from helical CT scan optimized with a large lung-size window scan in June 2019. Coronal CT reconstruction (A) highlighted the extent of colonic cystic wall pneumatosis, from the cecum to the proximal tract of the transverse colon (arrow). Axial CT scan (B) at the same level of the calcified peritoneal implant (black arrowhead), by applying a lung window, revealed a free retroperitoneal air bubble (white arrowhead) located between the abdominal aorta and the inferior vena cava.

No vascular occlusion nor portal-mesenteric pneumatosis were detectable. According to imaging findings, the radiologist proposed a diagnosis of large bowel PCI. Despite her good clinical status, the patient was referred to the emergency department for further examination. On admission, vital parameters were within normal limits. Laboratory tests showed mild normocytic normochromic anemia with a red blood cell count (RBCs) of 3.39 x 10^6/µl, and low hematocrit (31%) and hemoglobin level (10.5 g/dl). White blood cell count (WBC) was normal (5.07 x 10^3/µl) with a normal neutrophil count of 3.81 x 10^3/µl. However, lymphocytes (0.85 x 10^3/µl) and eosinophils (0.00 x 10^3/µl) levels were low. Electrolytes were balanced, except for sodium, slightly below the normal range (133 mmol/l). Other blood values were unremarkable as the laboratory tests showed platelets 321 x 10^3/µl; creatinine 0.86 mg/dL; glucose 106 mg/dL; total bilirubin 0.64 mg/dL; ALT 10 U/L; lipase 42 U/L; PCR 0.21 mg/dL. Urinalyses were unremarkable.

There were no signs of either peritonitis or sepsis, thus there was no indication for emergency laparotomy. Seven days later the patient was discharged with a final diagnosis of chemotherapy drug-induced PCI.

In July 2019, the patient started a new line treatment based on carboplatin monotherapy that is still ongoing.

A repeated CT after 6 months performed in an outpatient setting showed a reduction of the air collections in the intestinal wall, and the resolution of retroperitoneal free bubbles which were not detectable in the last CT after one year (Figure 3).

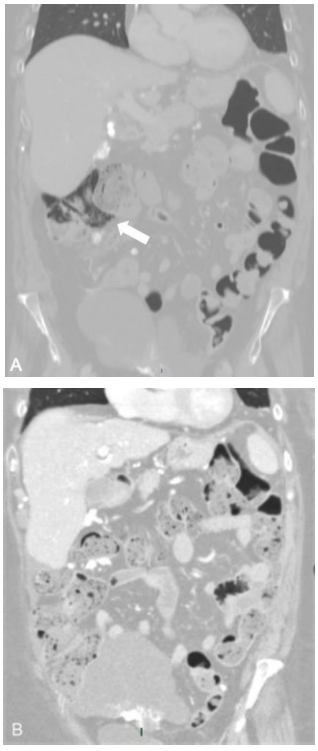

Figure 3. Coronal oblique multiplanar reconstruction from helical CT scan displayed in a large lung-size window. Coronal reconstruction in December 2019 (A) demonstrated a reduction in dimension and extent of the intramural colonic air collections (arrow) 6 months after the first detection of PCI, still maintaining a multilocular appearance. No intra-abdominal free gas signs were detected. A further CT scan in May 2020, a year later, showed the complete remission of pneumatosis (B).

4. Discussion

The first description of PCI is that of Du Vernoy in autopsy specimens in 1730, since then the scientific literature on the subject of PCI has flourished with an increasing number of case reports and studies (6). Nevertheless, PCI still represents a rare and poorly understood condition (7), the prevalence of which is difficult to establish, since most patients are asymptomatic. The gaseous cysts are typically located beneath the intestinal serosa and mucosa (4).

The most sensitive imaging technique for PI diagnosis is CT (3). Optic colonoscopy is frequently used to exclude underlying colonic lesions, and may reveal multiple small white cysts paired with a sub-atrophic mucosa, or larger cysts with a reddened over-lying mucosa (7,8).

On both abdominal radiographs and CT studies, PI can appear as hypodense linear, bubbly, or large cystic collections of gas in the intestinal wall, or as a combination of these patterns. Occasionally, bowel content mixed with intraluminal air trapped between mucosal folds may mimic PI; in this setting, CT may clarify any inconclusive or ambiguous finding.

The identification of this phenomenon may be facilitated using lung window settings and multiplanar reconstructions. Furthermore, CT is not merely more sensitive in the detection of PI but it can actually help to demonstrate the potential cause (2). The cystic variant of PI is more often benign; however, linear or bubble-like PI can be related to both benign and life- threatening causes (7).

The presence of additional CT features, such as thickened or paper-thin intestinal walls with altered enhancement, dilated intestinal loops, vessel occlusion, mesenteric stranding, ascites, and portal-mesenteric venous gas may suggest a potentially fatal cause (2,7), whereas the absence of these findings alongside clinical examination and laboratory data may result in conservative management (3).

PI can arise from pulmonary, systemic, or intestinal diseases and conditions. Several iatrogenic causes have been identified (3), including chemotherapy which may cause mucosal disruption and lymphoid depletion with subsequent passage of gas into the intestinal wall (7).

PI can occur in association with several chemotherapeutic drugs including cisplatin, cyclophosphamide, cytarabine, methotrexate, vincristine, doxorubicin, daunorubicin, etoposide, irinotecan, docetaxel (7, 9).

Notably, only one case has been previously described in association with Paclitaxel therapy (9). Here, for the first time, we describe a PCI diagnosis made after changing the chemotherapy regimen and which was assessed regularly by CT scan during its evolution .

It should be borne in mind that in these settings PI can be found even in asymptomatic patients, and encountered at routine surveillance imaging (10).

Reporting this finding correctly to the oncologist is important, because it may mean therapy discontinuation (3). Broadly speaking, the management of PI is guided by the severity of the clinical presentation, which can significantly vary on the basis of the underlying cause, ranging from benign to fulminant life-threatening conditions. The latter can entail high mortality if surgery is delayed (3).

Possible complications associated with PCI include bowel obstruction by internal or external compression when they increase in size, or intestinal perforation. Rarely, the rupture of gaseous cysts can produce pneumoperitoneum, reported in less than 3% of cases (7), or presence of air bubbles in the retroperitoneum or in the greater omentum; rarely portal-mesenteric venous gas may be observed as well in case of adsorption of the intramural gas by the draining mesenteric veins.

However, in oncological patients a conservative non-surgical approach is advised when there are no signs of peritonitis or sepsis (10).

In our case the patient was feeling well; therefore, careful observation and imaging follow-up were considered the most appropriate option.

In conclusion, PCI may be a complication of chemotherapy and sometimes may only require symptomatic treatment and close observation in order to promptly detect signs and symptoms suggestive of related complications.

References

1. Wang JH, Furlan A, Kaya D, Goshima S, Tublin M, Bae KT. Pneumatosis intestinalis versus pseudo-pneumatosis: review of CT findings and differentiation. Insights Imaging. 2011 Feb;2(1):85–92.
2. Ho LM, Paulson EK, Thompson WM. Pneumatosis Intestinalis in the Adult: Benign to Life-Threatening Causes. Am J Roentgenol. 2007 Jun;188(6):1604–13.
3. Khalil PN, Huber-Wagner S, Ladurner R, Kleespies A, Siebeck M, Mutschler W, et al. Natural history, clinical pattern, and surgical considerations of pneumatosis intestinalis. Eur J Med Res. 2009 Jun;14(6):231–9.
4. Azzaroli F, Turco L, Ceroni L, Galloni SS, Buonfiglioli F, Calvanese C, et al. Pneumatosis cystoides intestinalis. World J Gastroenterol. 2011 Nov;17(44):4932–6.
5. Eisenhauer EA, Therasse P, Bogaerts J, Schwartz LH, Sargent D, Ford R, et al. New response evaluation criteria in solid tumours: Revised RECIST guideline (version 1.1). Eur J Cancer. 2009 Jan;45(2):228–47.
6. Wang Y juan, Wang Y ming, Zheng Y min, Jiang H qing, Zhang J. Pneumatosis cystoides intestinalis: Six case reports and a review of the literature. BMC Gastroenterol. 2018;
7. Di Pietropaolo M, Trinci M, Giangregorio C, Galluzzo M, Miele V. Pneumatosis cystoides intestinalis: case report and review of literature. Clin J Gastroenterol. 2019 Jun;
8. Carnevale A, Bassi M, Ferrante Z, Rizzati R, Benea G, Giganti M. Computed tomographic colonography in the evaluation of a case of giant colonic diverticulum. BJR|case reports. 2017 Jun;3(2):20160101.
9. Guiu S, Ortega-Deballon P, Guiu B. Pneumatosis intestinalis and pneumoperitoneum during treatment by paclitaxel. Surgery. 2011;149(2):297–8.
10. Torrisi JM, Schwartz LH, Gollub MJ, Ginsberg MS, Bosl GJ, Hricak H. CT Findings of Chemotherapy-induced Toxicity: What Radiologists Need to Know about the Clinical and Radiologic Manifestations of Chemotherapy Toxicity. Radiology. 2011 Jan;258(1):41–56.

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