G Chir Vol. 38 - n. 5 - pp. 239-242
September-October 2017

clinical practice

Reversible pneumatosis cystoides intestinalis after liver transplantation

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SUMMARY: **Reversible pneumatosis cystoides intestinalis after liver transplantation.**

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Pneumatosis cystoides intestinalis (PCI) is a rare disease characterized by the presence of multiple gas-filled cysts within the submucosa or subserosa of the intestinal wall. We report a case of PCI in a 54-year-old man 4 weeks after liver transplantation due to hepatitis B virus (HBV)-associated liver cirrhosis presenting with illness, diarrhea and abdominal pain. CT scans revealed normal shape of the abdominal parenchymal organs and no intra-hepatic complication due to liver transplantation. Main abdominal venous and arterial vessels resulted patent. Colic loops appeared diffusely thickened for the presence of variable diameter air-filled cysts located within the bowel wall. The patient underwent conservative treatment and the CT control after 4 weeks showed a complete PCI resolution. PCI after adult liver transplantation is probably due to the pre-transplantation chemotherapy, immunosuppressive therapy and opportunistic enteric infections. Abdominal CT represents the gold standard technique for diagnosing PCI and for evaluating its extension and complications providing data on other abdominal pathologies. It appears as variable diameter gas-filled cysts located within the bowel wall and it is often associated with pneumo-peritoneum probably due to the rupture of subserosal cysts. PCI has a favorable outcome and requires conservative treatment. Major differential diagnosis includes intestinal pneumatosis due to bowel ischemia. PCI after liver transplantation represents an uncommon bowel disease with a favorable prognosis. CT represents the reference imaging technique for diagnosing the disease and evaluating the response to therapy which is usually conservative rather than surgical.

KEY WORDS: Colon - Pneumatosis cystoides intestinalis - Liver transplantation - Computed tomography - CT.

Introduction

Pneumatosis cystoides intestinalis (PCI) is a rare disease characterized by the presence of multiple gas-filled cysts within the submucosa or subserosa of the intestinal wall. It commonly affects patient with a mean age of 45.3 ± 15.6 years showing a male-to-female ratio of 2.4:1 and its most frequent location is reported to be the colon instead of the small bowel (rate of 1.3:1) (1-3).

Even if PCI etiology and pathogenesis are yet unknown, some theories have been proposed to explain this disease. They include mechanical theory which considers the intraluminal pressure as the main force causing intramural air bubble accumulation; the pulmonary theory which suggests that some lung diseases cause alveolar rupture, pneumo-mediastinum and air bubble migration to the bowel wall through the aorta and mesenteric vessels; the bacterial theory which suggests that intramural air bubbles are produced by gas-forming bacteria entering the mucosal layer (3-5). The bacterial theory could explain the occurrence of PCI in immuno-compromised patients who have undergone organ transplantation. In particular, it has been rarely reported in adult liver transplant recipients with a prevalence of less than 1% (6-9).

Among all diagnostic tools for abdominal imaging, Computed tomography (CT) represents the gold standard technique for evaluating both abdominal organs and bowel loops also providing a guide for interventional diagnostic procedures in selected cases (10-13).

In the field of bowel diseases, CT can accurately iden-

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tify PCI findings allowing to perform a differential diagnosis with intestinal pneumatosis due to bowel ischemia and to carry out the correct therapeutic approach (1, 14).

We report an uncommon case of PCI occurring after adult liver transplantation.

Case report

A 54-year-old man was admitted to our hospital 4 weeks after liver transplantation due to hepatitis B virus (HBV)-associated liver cirrhosis. The patient complained of general feelings of illness, diarrhea and abdominal pain. Physical examination revealed only abdominal distension and laboratory findings were within normal ranges. Abdomen radiograph showed grape-like radiolucency located along the wall of the entire colon. Therefore, CT examination was mandatory.

The patient was scanned from diaphragmatic dome to the pubic symphysis, before and after intravenous injection of 1.5 mL/kg of Iopamidol (Iomeron 400; Bracco, Milan; Italy) at 3.5 mL/s through the ante-cubital vein with an automatic power injector.

A 320-row CT scanner was used (detector collimation 0.5 mm, increment 0.5, 120/250 kVp/mAs). CT acquisition was performed with a biphasic technique during the arterial and portal venous phases after the intravenous injection of contrast material. An additional delayed phase was obtained from the diaphragmatic dome to the pubic symphysis.

All CT data were transferred to a workstation (HP XW 8600) equipped with dedicated software (Vitrea FX 2.1, Vital Images, Minneapolis, Minnesota, US) for image reconstructions.

CT scans revealed normal shape of abdominal parenchymal organs and no intra-hepatic complication due to transplantation was found. Main abdominal venous and arterial vessels resulted patent. Colic loops appeared diffusely thickened for the presence of variable diameter air-filled cysts located within the bowel wall (Figure 1). Pneumo-peritoneum and a subtle fluid pelvic collection were associated.

The patient underwent conservative treatment consisting of parenteral nutrition, fluid supplementation, steroid stop and antibiotics. CT examination performed after 4 weeks showed a complete resolution of the pneumo-peritoneum and of the colic findings with a normal shape and thickness of the colic loops.

Discussion

Few cases of PCI after adult liver transplantation have been reported in the medical literature, probably due to the pre-transplantation chemotherapy, immunosuppressive therapy and opportunistic enteric infections. The bowel wall involvement commonly causes gastrointestinal symptoms mainly represented by diarrhea, abdominal pain and distension, bloody stool, constipation and tenesmus. However, in some cases the disease is asymptomatic and recognized incidentally on routine abdomen radiographs or CT scans (1-5, 14).

The laboratory findings do not reveal abnormal findings except for mildly elevated C-reactive protein. Serial blood tests are positive for cytomegalovirus (CMV) antigenaemia in some cases (1, 6, 7, 14).

PCI is reported to occur from few weeks to many months after liver transplantation and its diagnosis relies on abdomen radiography, CT and colonoscopy. Abdomen radiography shows grape-like radiolucency along the bowel wall. Abdominal CT represents the gold standard technique for diagnosing PCI and for evaluating its extension and complications providing data on other abdominal pathologies. On CT images, PCI appears as variable diameter gas-filled cysts located within the bowel wall and it is often associated with pneumoperitoneum probably due to the rupture of subserosal cysts. Colonoscopy reveals multiple round and smooth-surfaced elevated lesions appearing as hyperechoic lesions with acoustic shadows when endoscopic ultrasonography is performed (1, 6, 7, 14).

The majority of patients affected by PCI undergo conservative treatment consisting of bowel rest, parenteral nutrition, fluid and electrolyte supplementation, steroid or immunosuppressant stop. It ensures gas disappearance on CT scans within few weeks. On the other hand, the PCI surgical treatment is associated with high mortality rate ranging between 33 and 44% (1, 14).

Major differential diagnosis includes intestinal pneumatosis due to bowel ischemia which represents an abdominal emergency occurring in nearly 1% of patients presenting with acute abdomen. However, ischemic parietal pneumatosis is characterized by a typical linear or curved shape of air within bowel wall (1, 14-17).

Conclusions

PCI after liver transplantation represents an uncommon bowel disease with a favorable prognosis and occurring in specific clinical settings. CT represents the reference imaging technique for diagnosing the disease and for performing the relevant treatment which is conservative rather than surgical.

Conflicts of interest

The authors declare no conflicts of interest.

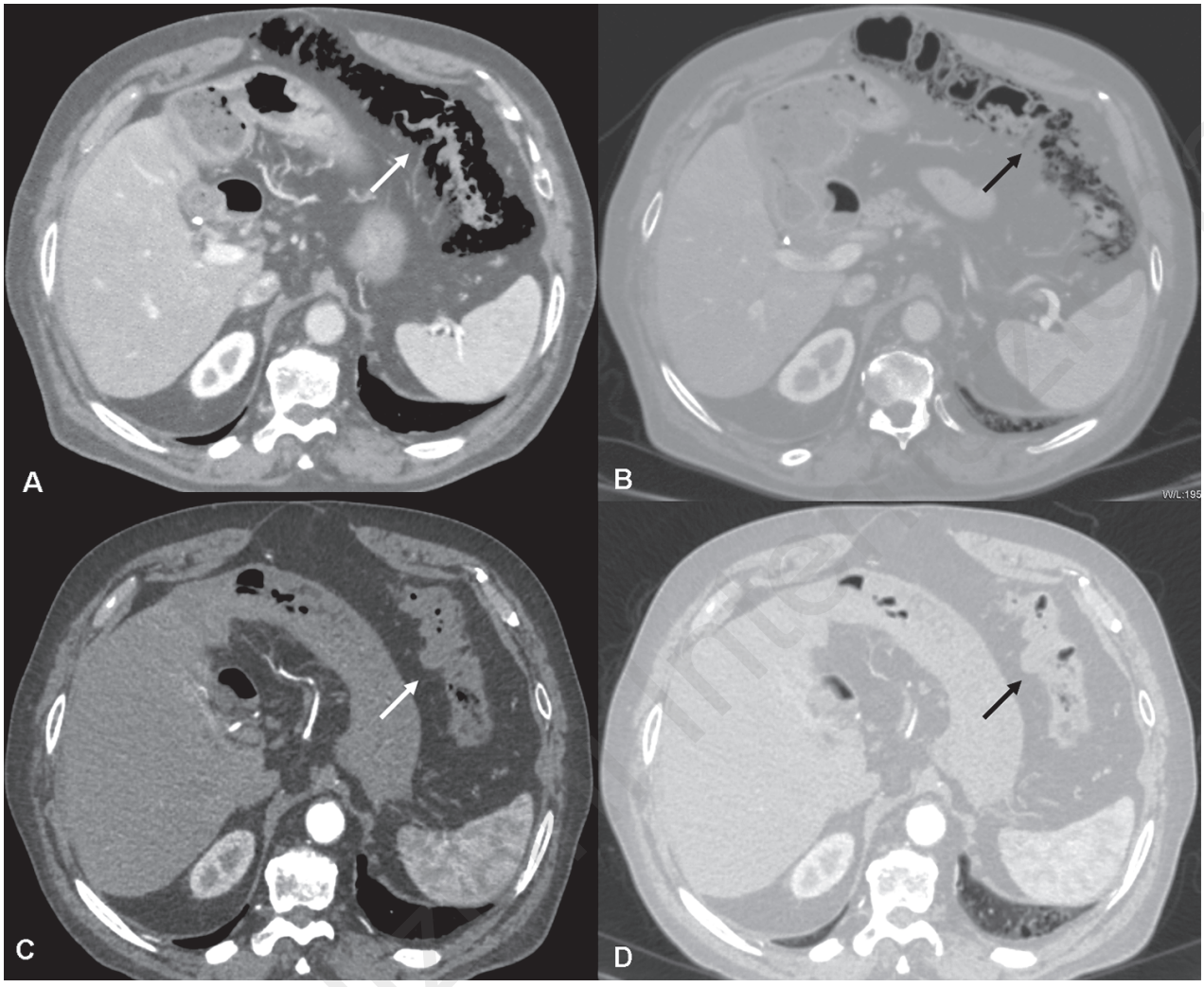

Figure 1 - Transverse CT scans (A, soft tissue window; B, lung parenchyma window) show pneumatosis cystiodes intestinalis of the colon represented by multiple gas-filled intramural cysts (arrow) in an adult liver recipient. The CT control after 4 weeks of conservative therapy shows a complete resolution of the disease (arrow) (C, soft tissue window; D, lung parenchyma window).

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