Pneumatosis cystoides intestinalis of small bowel and cholecysto-duodenal fistula in stenosing peptic ulcer revealed by pneumoperitoneum

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Citation

Abstract
Pneumatosis of small bowel is a rare disease. Its association with a cholecysto-duodenal fistula is not often reported. This is the case of a 30-year-old man with a past medical history of duodenal ulcer, who presented in the emergency department for non-specific abdominal pain. Abdominal X-ray revealed a pneumoperitoneum. Computed tomography showed a pneumoperitoneum, presence of air in the biliary tract and gas in the small bowel wall. Gastro-intestinal opacification revealed a cholecysto-duodenal fistula in a stenosing duodenal ulcer associated with pneumatosis cystoides intestinalis. The patient went on to have a truncal vagotomy and gastrojejunal anastomosis. A cholecystectomy and a duodenal repair were also performed. Computed tomography and a gastrograffin study are very important for the diagnosis of pneumatosis cystoides intestinalis with cholecysto-duodenal fistula in stenosing ulcer.

INTRODUCTION
Pneumatosis cystoides intestinalis (PCI) of small bowel is a rare disease which presents with gas bubbles in the small bowel wall on radiological studies [1]. Its association with cholecysto-duodenal fistula is not often described and gets diagnostic and therapeutic interest. We report a case of a young patient, in whom the discovery of a pneumoperitoneum led to the diagnosis of PCI associated with a cholecysto-duodenal fistula on a stenosing peptic ulcer.

CASE REPORT
A 30 years old male presented in the emergency department in April 2004 for right hypochondrial pain associated with vomiting over one week. At his examination, the patient presented in a good general condition with a temperature of 100.4°F; there was no sign of peritoneal irritation. The rest of the examination and biological tests were normal. Standard abdominal X-ray noted a pneumoperitoneum. The abdominal computed tomography with gastrointestinal opacification found air in the intrahepatic biliary tract associated with a pneumoperitoneum (figure 1) and cysts on the small bowel wall which were in the supracolic compartment, between the liver and the abdominal wall (figure 2). The diagnosis of PCI associated with a biliodigestive fistula was made and the patient was subjected to a gastrograffin study. The gastro-intestinal opacification found a cholecysto-duodenal fistula on a stenosing duodenal peptic ulcer with bullous images of PCI (figure 3). The patient underwent a laparotomy which led to the discovery of cystic lesions in the terminal part of the jejunum and the ileum (figure 4). Much of the small bowel was located between the liver, the abdominal wall and the diaphragm. There was a stenosing duodenal peptic ulcer and an inflammatory aspect of the pyloro-duodenal area which was the seat of the cholecysto-duodenal fistula. A truncal vagotomy with a gastro-jejunostomy was performed. Cholecystectomy and closing of the duodenal fistula were carried out. The postoperative recovery was uncomplicated. Histological analysis of the gallbladder revealed a chronic cholecystitis. After a follow-up of five years, the patient has not presented with symptoms and has no complaints of abdominal pain.
Figure 1
Figure 1. Computed tomography: Pneumobilia and pneumoperitoneum. Note gastric distension due to duodenal stricture.

DISCUSSION
PCI is characterized by the presence of gas bubbles in the gastrointestinal wall. It is a rare disease; however, its frequency is underestimated because of asymptomatic forms [2]. Several pathophysiological theories are put forth. The theory of the lung origin of the bubbles refers to a reduction of the gas absorption capacity and their perivascular migration from alveolar failure during chronic obstructive pulmonary disease [3]. The mechanical theory explains injury by high-pressure penetration of air in the intestinal wall to a pre-existent mucosa breach [4]. The bacterial theory explains the presence of gas in the intestine wall by rapidly multiplying bacteria in the sub-mucosa [5]. PCI is asymptomatic in most cases [6] but complications are described, like intussusception, intestinal necrosis and perforations of cysts [2, 3, 4]. The diagnosis of PCI is made by standard abdominal X-ray with the presence of vacuolated images in the bowel wall grouped into clusters of grapes or in aggregates or by the presence of a pneumoperitoneum in a patient who does not have a gastrointestinal perforation [7]. Computed tomography shows cystic lesions lining the edge of the bowel wall which is the most sensitive sign. It specifies the primary (in the absence of an abdominal cause) or secondary character [8, 9]. This disease is idiopathic in 15% of the cases [4]. Also, it is associated with obstructive bronchitis, tumors or gastrointestinal strictures [1, 10, 11]. A peptic ulcer stricture of the duodenal bulb is encountered in 50% of cases of PCI [12], as in our observation; it is exceptionally complicated by a cholecysto-duodenal fistula. In our practice, this is the first PCI – cholecysto-duodenal fistula association which complicated a stenosing peptic ulcer. In our observation, the discovery of a pneumoperitoneum at standard abdominal X-ray on a non-surgical painful abdomen was decisive for the performance of an abdominal computed tomography. CT scan showed pneumobilia in the absence of previous biliary surgery associated with images of gas bubbles on the edge of the small-bowel wall and lead to the diagnosis of PCI associated to a bilo-gastrointestinal fistula. The water-soluble swallow confirmed the cholecysto-duodenal fistula on a stenosing peptic ulcer which was also confirmed during laparotomy. The surgical treatment of PCI is indicated only for complicated cases. In our observation, bilateral truncal vagotomy and gastro-jejunostomy were done to treat the duodenal ulcer and its complications. In the absence of complications, the treatment consists in oxygenotherapy and antibiotherapy [13]. In cases of cholecysto-duodenal fistula, cholecystectomy is strongly recommended to avoid the appearance of chronic cholecystitis on a gallbladder made hypotonic [14].

CONCLUSION
Pneumatosis cystoides intestinalis associated with a cholecysto-duodenal fistula is rare. Surgical treatment of such an association needs peptic ulcer stricture treatment and cholecystectomy. No action is carried out on PCI in the absence of complications. This observation highlights the importance of the CT scan in the diagnosis.

References
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