Gallstone Ileus and Crohn's Disease without Biliary-Enteric Fistula:

Report of a Unique Case

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Abstract

Gallstone ileus is an uncommon cause of small bowel obstruction, accounting for fewer than 3% of laparotomies for intestinal obstruction. Patients with long-standing Crohn's disease have an increased risk of developing gallstone disease. However, gallstone ileus is not common in these patients. We report the case of a 70-year-old female with Crohn's disease who presented with gallstone ileus, and present a review of the literature. We discuss the association between gallstone ileus and Crohn's disease, and the treatment options for these patients. We emphasize the importance of including gallstone ileus in the differential diagnosis in patients presenting with intestinal obstruction, especially patients with long-standing Crohn's disease. We advocate the early utilization of computerized tomography to confirm the diagnosis, and prompt early surgical intervention. **Key Words:** Crohn's disease, gallstone ileus.

Introduction

GALLSTONE ILEUS is an uncommon cause of small bowel obstruction. It accounts for fewer than 3% of laparotomies for intestinal obstruction. Most patients are over the age of 65 and female (1). A cholecystoduodenal fistula is found in the majority of cases. The stone is typically of large size and the usual site of impaction is the distal ileum (2). Treatment is laparotomy and relief of obstruction by enterolithotomy. Patients undergoing surgery for gallstone ileus have reported mortality and morbidity rates of 11.7% and 32%, respectively (1).

Patients with long-standing Crohn's disease have an increased risk of gallbladder disease (3), but there are few reports of gallstone ileus in these patients. The increased risk of gallbladder disease is attributed to the disturbance in the enterohepatic circulation of bile salts secondary to disease of the terminal ileum and a resultant change in cholesterol solubility leading to the increased formation of gallstones (4).

We report a case of a 70-year-old female with Crohn's disease who presented with gallstone ileus requiring surgical intervention, and present a review of the literature. We discuss the association between gallstone ileus and Crohn's disease, and the treatment options for these patients.

Case

A 70-year-old female with a 35-year history of Crohn's disease involving the terminal ileum was admitted for small bowel obstruction. Her past medical history was significant for breast cancer requiring left mastectomy at the age of 49, and mild chronic bronchitis. Medications were beclomethasone inhalers and diphenoxylate. The diagnosis of Crohn's disease was first made at the age of 35, at exploratory laparotomy in which the diseased terminal ileum was left *in situ* and an ileotransverse colon side-to-side anastomosis was performed. At the age of 42, the patient had resection of the bypassed segment for intractable

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Crohn's disease, and the previous ileocolic anastomosis was left intact. After the second operation, the patient remained relatively asymptomatic and complained only of occasional episodes of intermittent abdominal pain. On the day of admission, the patient developed increasing abdominal pain and distention, nausea and bilious vomiting. On physical examination, the abdomen was distended and tympanitic without signs of peritonitis. An obstructive series was consistent with small bowel obstruction and an opacified density in the left abdomen was noted (Fig. 1). Computerized tomographic scan (CT scan) of the abdomen showed this opacified mass to be intraluminal and highly suspicious for a gallstone (Fig. 2). Sonogram of the abdomen revealed sludge in the gallbladder, without biliary dilatation. The patient was rehydrated and taken to the operating room 24 hours after admission. Exploratory laparotomy revealed distended loops of small bowel and a clear transition zone from dilated to collapsed bowel just proximal to the previous ileotransverse colon anastomosis. A 3.5 cm stone was palpated at this site. The neoterminal ileum was diseased and strictured secondary to Crohn's disease. The neoterminal ileum, along with adjacent colon, was resected and a primary anastomosis was performed (Fig. 3). Adhesions to the gallbladder were taken down, and the gallbladder was visualized and palpated. There were no signs of inflammation, there were no palpable stones, and there was no fistulous communication between the gastrointestinal tract and the gallbladder. No other stones could be palpated in the duodenum, small bowel or large bowel. The patient's postoperative course was uneventful and she was discharged on the ninth postoperative day. Polarized microscopic analysis of the stone (LabCorp, Burlington, NC) showed that calcium bilirubinate was the main component, comprising 80% of the

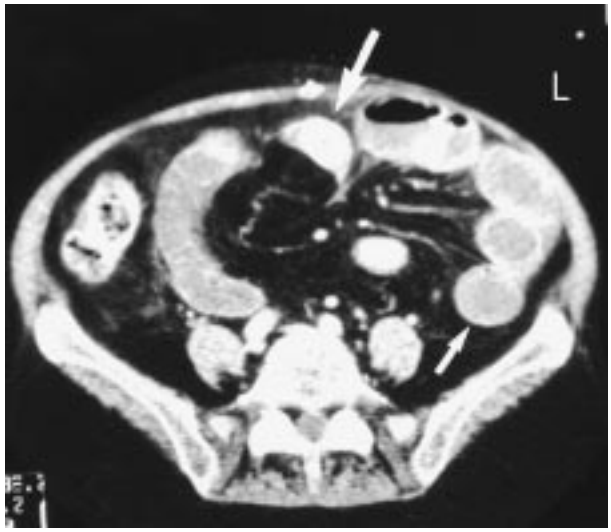

Fig. 2. Computerized tomographic scan of the abdomen demonstrating dilated loops of small bowel (small arrow) and an intra-luminal calcified mass (large arrow). The intrahepatic and extrahepatic bile ducts were normal.

Fig. 1. Plain abdominal radiograph showing dilated loops of small bowel (arrow) and a calcified mass in the left abdomen (arrowhead).

Fig. 3. Photograph of the surgical specimen showing the stone (arrow) impacted in a segment of diseased and strictured small bowel (arrowheads).

stone and all of the core. Gross and microscopic examination of the distal ileum confirmed Crohn's disease.

Discussion

Gallstone ileus accounts for fewer than 3% of laparotomies for small bowel obstruction (2). Most patients are over the age of 65 with a male to female ratio of 1:5 to 1:10. Biliary symptoms are common, but the diagnosis is made preoperatively in less than 50% of cases (5). The most common pathology is a cholecystoduodenal fistula, although fistulous connections have been reported to the jejunum, colon and stomach (6). A few case reports of intestinal obstruction caused by stones without a fistulous communication have been reported. As was previously suggested, a possible explanation can be the migration of a small stone through the papilla of Vater and "*in-situ* growth" in strictured small bowel (7).

Because its luminal diameter is small, the ileum is the most frequent site of impaction, but stones have been found impacted from the stomach to the sigmoid colon (8). Symptoms are usually intermittent and consistent with partial intestinal obstruction, the so-called "tumbling phenomenon." A flat plate of the abdomen will reveal small bowel obstruction, air in the intrahepatic ducts, and a mobile opacified mass in 25% of patients (9). In the present case, abdominal radiographs were consistent with small bowel obstruction and a calcified mass was found, but air in the intrahepatic ducts was not noted. Treatment is laparotomy with relief of the obstruction. Cholecystectomy and repair of the fistula, when present, are subjects of controversy.

Crohn's disease is strongly associated with gallstone disease. The prevalence is especially high in patients with Crohn's disease of the terminal ileum and in patients after ileal resection (3, 10). Cholesterol is the main component of most cases of gallstones in patients with Crohn's disease. The pathogenesis is attributed to the disturbance in the enterohepatic circulation caused by decreased absorption of bile salts, secondary to Crohn's disease, or because of resection of the terminal ileum. The diminished pool of bile salts changes the solubility of cholesterol in bile, causing cholesterol to precipitate (4). Ileal dysmotility has also been suggested as a cause. Lapidus, however, found that the concentration of bilirubin in bile in patients with Crohn's disease after ileal resection is 45–50% higher than in healthy controls (11) and suggested that patients with Crohn's disease develop pigment stones rather than cholesterol gallstones. Increased concentration of bilirubin in a segment of the small bowel may enhance "*in-situ* growth" of a small pigment stone. This can be a possible explanation for the finding of a stone comprised mainly of calcium bilirubinate in a patient without a fistulous communication to the gallbladder, as was the case in this patient.

An association between Crohn's disease and gallstone ileus is rare (12, 13); only three cases have been reported. The mean age was 60.3 years (50–67 years), with a mean duration of disease of 13.7 years (4–20 years). Mean stone size was 2.5 cm (1.5–3 cm), considerably smaller than reported for patients without Crohn's disease (mean 4.5 cm, range 2–9 cm). This is presumably due to the already narrowed small bowel lumen secondary to the fibrostenotic Crohn's disease. Accordingly, the patient we present belongs to an older group of patients, with long duration of disease, who presented with intestinal obstruction caused by a small stone.

Of the three known cases in the literature of Crohn's disease complicated by gallstone ileus, only one had biliary symptoms. The diagnosis of gallbladder disease in patients with Crohn's disease can be missed as a result of the predominance of intestinal symptoms in these patients. In a review of 1001 patients with gallstone ileus, preoperative diagnosis was made in only 43% (1). Only one of the three patients with gallstone ileus and Crohn's disease was diagnosed preoperatively. In patients with Crohn's disease presenting with uncomplicated small bowel obstruction, medical therapy is generally the first line of treatment, because much of the luminal narrowing may be related to inflammation which may be treated pharmacologically. Also, unlike adhesive obstruction, ischemia and gangrene of the bowel is very rare in this group of patients. As was previously reported, the addition of a CT scan in this case to confirm the diagnosis prompted early surgical intervention (14).

Laparotomy revealed the stone to be impacted in a strictured segment of diseased small bowel. In patients with Crohn's disease, obstruction at a strictured and diseased segment of bowel, even with a small stone, is not surprising. Furthermore, with the combination of lithogenic bile and small bowel narrowing due to Crohn's disease, a higher incidence of gallstone ileus in patients with Crohn's disease might be expected.

The operative approach to gallstone ileus is enterotomy, extraction of the stone and a careful search for more stones and a biliary-enteric fistula (1). We were faced with a more complex prob-

lem due to the fact that the stone was impacted in diseased bowel. The approach in such a patient should be different and include a procedure that resects, bypasses, or widens the strictured segment of small bowel. Our patient, as well as the three previously reported cases, underwent ileocolic resection and restoration of bowel continuity.

Conclusion

We present a case of a 70-year-old female with long-standing Crohn's disease who presented with intestinal obstruction secondary to gallstone ileus. Early surgical intervention was prompted by the use of CT scan to confirm this uncommon diagnosis. Intestinal obstruction was caused by a small gallstone impacted in strictured ileum secondary to Crohn's disease. No bilio-enteric fistula was found. The differential diagnosis of intestinal obstruction should always include gallstone ileus, especially for patients with long-standing Crohn's disease. The operative approach is resection of the segment of bowel with restoration of bowel continuity. A search for a fistulous communication between the biliary tract and the intestine is also necessary.

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