1. Introduction

Gallstone ileus is a rare complication caused by repeated episodes of cholecystitis, or rarely malignancy, with a predominance in elderly patients with comorbidities.[1,2] Poor appetite and recurrent vomiting caused by ileus can worsen existing poor health conditions in the elderly.[3] In patients of advanced age with associated comorbidities and a delayed presentation, the mortality rate for gallstone ileus can be as high as 18%.[1] Early diagnosis followed by suitable and timely surgical intervention are considered 2 predictors of improved prognosis.[1–3]

Gallstone ileus displaying the typical Rigler triad and an occult second ectopic stone

A case report

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Abstract

Rationale: Gallstone ileus is a rare complication of cholecystolithiasis. It has a female predominance and can result in high mortality rates.

Patient concerns: A 71-year-old woman complaining of recurrent vomiting and vague epigastralgia for > 2 weeks presented to our department.

Diagnosis: Based on her physical examination, laboratory test results and radiographic findings (the typical Rigler’s triad), she was diagnosed with gallstone ileus caused by multiple ectopic stones.

Interventions: After correction of electrolyte imbalances through parenteral nutrition and fluid management, the patient’s condition improved and she underwent enterolithotomy. A 5.3-cm stone located 40 cm from the ligament of Treitz was extracted, and a second ectopic stone, not detected on any imaging modality, was found during laparotomy. Given her comorbidities and overall poor condition, cholecystectomy and fistula repair were not performed.

Outcomes: The patient gradually recovered postoperatively.

Lessons: Clinical symptoms including epigastralgia with nausea and vomiting, and abdominal CT findings of Rigler’s triad (ectopic gallstone, bowel obstruction, and pneumobilia) may lead to early diagnosis of gallstone ileus and enterolithotomy may be the preferred treatment for this condition in the elderly. Laparotomy should involve a systematic and meticulous search for the presence of additional ectopic enteric stones.

Abbreviations: CRP = C-reactive protein, CT = computed tomography, ESR = erythrocyte sedimentation rate.

Keywords: cholecystoenteric fistula, enterolithotomy, gallstone ileus, multiple stones, Rigler triad

2. Case report

A 71-year-old woman presented to the Hepato-Bilio-Pancreatic Department complaining of recurrent attacks of vomiting bile-stained gastric contents with palpitations, weakness, difficulty in defecation, and loss of appetite. She had sought medical help at a local hospital during the initial stage of this episode. Unfortunately, nonsurgical management did not relieve her symptoms and her overall condition worsened. About 1 year before this episode, a gallbladder stone had been detected on ultrasound following an episode of right upper quadrant pain. She had been diagnosed with type 2 diabetes mellitus for > 20 years and had an oophorectomy 40 years prior.

Rigler triad, a pathognomonic radiographic finding is not routinely observed on plain x-ray or ultrasound but is identified on CT scan with much higher accuracy.[4] Given the rare occurrence of gallstone ileus, a clear consensus on the optimal surgical intervention has not been reached.[1,3] Despite the lack of consensus, careful and thorough examination of the entire small bowel during laparotomy has been generally accepted as standard practice, as multiple stones can occur in 5% to 25% of cases of this disease.[1,2]

In this report, we present a case of surgically treated gallstone ileus with findings of the typical Rigler triad as seen on 3-dimensional reconstruction images. Moreover, after removal of the primary stone (5.3 cm in diameter), an ectopic stone not detected on any imaging examination was found during laparotomy in the same patient.
On physical examination she was afebrile (36.9°C), normotensive (92/60 mm Hg), and without signs of jaundice. Abdominal examination revealed remarkable abdominal distention with sensitivity in the periumbilical area. Murphy sign was negative. Laboratory test results were as follows: complete blood count revealed anemia (red blood cell count: 2.98 × 10^{12} L^{-1}, reference range: 4.3–5.8 × 10^{12} L^{-1}); hemoglobin 87 g/L, reference range 130–175 g/L); blood biochemical studies revealed slightly decreased total bilirubin (4.6 μmol/L, reference range 5.0–28 μmol/L), significantly decreased albumin (19.2 g/L, reference range 30–50 g/L), and decreased calcium (1.69 mmol/L, reference range 2.1–2.7 mmol/L). Inflammatory factors including leukocyte count, erythrocyte sedimentation rate (ESR), and C-reactive protein (CRP) were normal.

Abdominal sonography revealed a slightly thickened gallbladder wall with air in the gallbladder. The plain abdominal film depicted air-fluid levels and dilatation of the loops of the small bowel (Fig. 1, black arrows). To identify the obstructive cause, abdominal computed tomography (CT) was performed with a helical unit (slice thickness 5 mm, reconstruction interval 2 mm, Somatom Definition Flash, Siemens Healthcare, Forchheim, Germany). Transverse, non-enhanced CT images revealed a round intraluminal lesion measuring 3.4 × 3.8 cm with central hypoattenuation in the jejunum (Fig. 2A). The 3-dimensional reconstruction images revealed the typical Rigler triad of ectopic gallstone (Fig. 2), bowel obstruction (Fig. 2B, white arrow), and pneumobilia (Fig. 3). Coronal abdominal CT images also demonstrated a cholecystoduodenal fistula between the gallbladder and the descending duodenum (Fig. 3B, white arrow). These radiographic findings, particularly the abdominal CT images revealing the typical Rigler triad, strongly supported the diagnosis of gallstone ileus and cholecystenteric fistula.

The initial management for this elderly woman was to improve her poor general condition and correct her electrolyte imbalances through parenteral nutrition and fluid management. In addition to the treatment aimed at improving her overall condition, we expected that the stone could pass through the ileocecal valve if we prescribed oral medical paraffin oil. However, after becoming aware of the size of the stone, conservative treatment was abandoned and more aggressive treatments were considered. We did not attempt endoscopic management of the stone that was in the intestinal lumen. Ultimately, after improvement in her general condition, the patient underwent enterolithotomy. During this procedure, we removed a 5.3-cm ectopic stone (Fig. 4) that was located 40 cm from the ligament of Trettz in the proximal jejunal lumen. Interestingly, while inspecting the entire length of the small and large intestines, we found and extracted another small stone measuring 2.0 cm in diameter (Fig. 4) in the distal ileal lumen. This stone had not previously been detected on any imaging modalities. Given the severe adhesions in the abdominal cavity, the comorbidities, and the poor overall condition of this patient, cholecystectomy and fistula repair were not performed. Despite development of a severe infection in the surgical incision and persistently poor overall health, the patient gradually recovered and was discharged 22 days postoperatively.

This case report was approved by the Ethics Committee of West China Hospital of Sichuan University, Chengdu, China, and written informed consent was obtained.

3. Discussion

Gallstone ileus, a rare complication of cholecystolithiasis occurring in <1% of patients, is the cause of 1% to 4% of cases of small bowel obstruction. [1–3] As a pathogenesis of gallstone ileus, cholecystoenteric fistula caused by recurrent cholecystitis is identified most commonly in elderly women with comorbidities. [1,2] Gallstones pass through fistulas into the duodenum, stomach, colon, or intestine, and most of the them are excreted. [2,3] Ectopic stones obstructing the intestinal lumen can lead to ileus. The atypical presentation, along with the low incidence of the disease in elderly patients, decreased doctors’ vigilance upon first presentation of these patients. [1,2] Consequently, there was a delay in correctly diagnosing these patients’ illness, leading to a worsening general condition. Although advances in anesthesia and perioperative management can improve patients’ prognoses dramatically, the mortality and morbidity remain considerable. [1–3] To the best of our knowledge, the presence of the typical Rigler triad visualized on ultrasound and plain x-ray is only obvious in a small number of cases of this disease (11.11% and 14.81%, respectively). Additionally, the incidence of a second, or multiple additional, ectopic stones was as low as 5% for this relatively rare disease. In this report, we presented the typical Rigler triad based on 3-dimensional reconstruction CT images. We also noted the presence of a second ectopic stone that was not detected on any imaging modality in the same patient.

Abdominal pain and vomiting are constant but nonspecific features for gallstone ileus, and >50% of patients present with features of intestinal obstruction. Other nonspecific signs include dehydration, upper gastrointestinal hemorrhage, abdominal tenderness, and pyrexia. [1–3] In summary, the clinical manifestation of gallstone ileus is variable and nonspecific and attention should be paid to any history of recurrent episodes of acute cholecystitis, which occurred in approximately half of these cases. [1,2] In this case, the patient’s main clinical manifestation was recurrent vomiting without obvious abdominal pain.

Imaging studies play a vital part in the early diagnosis of gallstone ileus. Rigler triad, described as signs of partial or complete intestinal obstruction, air or contrast medium in the biliary tree, and ectopic gallstone is the specific imaging finding.
for this disease, and can be detected on x-ray, ultrasound, and CT. Unfortunately, only 13.81% of gallstone ileus patients present with the typical Rigler triad on plain x-ray. Ultrasound is useful in detecting cholecystitis and pneumobilia, but not valuable in diagnosing the intestinal situation or the location of an ectopic gallstone. Presently, CT has been proven to be the most valuable diagnostic modality for gallstone ileus, having optimal sensitivity, specificity, and accuracy (93%, 100%, and 99%, respectively). In addition to the detection of Rigler triad, CT can also provide vital information such as fistula location, or severe inflammation of the gallbladder and its surrounding tissues. This type of information is useful for early diagnosis and optimal therapies. MRI could be used for confirmation of findings before treatment. Unfortunately, some smaller and less calcified stones may still be missed by this imaging format, as described in this case.

The debate over which surgical procedure should be undertaken has lasted for years due to the low incidence of this disease. The primary conflicts involve the decision whether to perform cholecystoenteric fistula repair during the initial procedure. A review of 1001 cases concluded that simple enterolithotomy was both safe and effective in managing a patient with gallstone ileus. A review of recent cases of patients with gallstone ileus who underwent surgical treatment concluded that even after adjusting for patient and hospital factors,
compared with simple enterolithotomy, fistula repair performed as the initial procedure was associated not only with higher mortality rates, but also with longer hospital stays and greater total treatment expense. In our case, although we solely performed enterolithotomy, the patient still recovered slowly due to a severe infection of the abdominal incision and hospitalization for 22 days postoperatively. Indeed, a more aggressive surgical procedure might cause even greater difficulties with postoperative recovery.

The presence of second or multiple stones may be as low as 5% in patients with gallstone ileus. However, the inability to detect these stones might result in extremely poor outcomes. Indeed, even 1 stone remaining in the colon could lead to a lethal outcome. CT has been proven to be the most useful radiographic modality for diagnosis and preoperative evaluation. However, in the present case, after meticulous examination of the small and large bowel, we found a second ectopic stone in the distal ileum that was not detected by any of the preoperative imaging modalities, including 3-dimensional reconstruction CT images.

4. Conclusions

Gallstone ileus is a rare but severe complication of cholecystitis. Nonspecific symptoms and a predilection in the elderly caused a delay in diagnosis. The presence of ileus, electrolyte disturbances, hypoproteinemia, anemia, delay in diagnosis, and comorbidities, all contributed to the patient’s poor overall condition. Early surgical intervention following early diagnosis of gallstone ileus is of vital importance. CT, which was demonstrated to be a valuable test in early diagnosis, should be performed in any elderly patient complaining of abdominal pain and recurrent vomiting with a history of cholecystolithiasis. Actually, enterolithotomy was proven to be the most beneficial operation for patients in poor overall health, and laparotomy should involve a systematic and meticulous search for the presence of further enteric stones.

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