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Case report and review of the literature



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Viola Zulian, Giorgio Vasquez*, Carlo V. Feo

Sezione di Clinica Chirurgica, Dipartimento di Scienze Chirurgiche, Radiologiche e Anestesiologiche at the Università di Ferrara, Italy
*Modulo di Chirurgia d'Urgenza, Dipartimento Chirurgico, Azienda Ospedaliero-Universitaria "S. Anna", Ferrara, Italy

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Gallstone ileus is a rare complication of cholelithiasis. It accounts for 25% of nonstrangulated small bowel obstructions in patients over the age of 65 years. The morbidity and mortality rate of gallstone ileus remains very high, partly because of misdiagnosis or delayed diagnosis. The two surgical options are: a) enterolithotomy with removal of impacted stone, cholecystectomy, and fistula repair at the same surgical operation (i.e., "one-stage" procedure) and b) enterolithotomy with stone extraction followed or not by elective biliary surgery. The latter is the most popular surgical approach, whereas enterolithotomy combined with cholecistectomy and fistulectomy is indicated only in selected cases. In this article, a case of biliary ileus with unusual presentation treated by enterolithotomy alone with long term follow up is described, and the literature on this subject is reviewed and discussed.

KEY WORDS: Aged, Biliary Fistula, Cholelithiasis, Gallstones, Ileus.

Introduction

Gallstone ileus is an uncommon complication of cholelithiasis (0.3-0.5%) which mainly presents in elderly patients, accounting for 1% to 4% of all small bowel obstructions¹. Probably, longevity in Western countries has increased the prevalence of gallstone ileus, which represents 25% of nonstrangulated small bowel obstruction in patients over 65 year old². Apparently, the disease was first described by Erasmus Bartolin, a Danish physician, in 1654, although Courvoisier published the first series of 131 patients in 1890³. Gallstone ileus may present at a wide range of age, but the mean age reported in the largest series is 72 years (range, 13-97 years)⁴. It occurs more frequently in women than in men in a ratio ranging between 4:1 and 16:12. The pathogenesis involves large gallstones causing extensive inflamma-

tion and adhesion of the gallbladder to the adjacent gastrointestinal tract, followed by pressure and ischemia yielding to erosion of the stone into the bowel wall (i.e., cholecystoenteric fistula), with subsequent mechanical bowel obstruction. However, cancers as well as penetrating ulcers have been reported to cause gallstone ileus⁵. Most commonly, a cholecystoduodenal fistula occurs (69%), but the fistula may develop between the gallbladder and the ileus (20.3%), colon (8.5%) or stomach (1.6%)⁶. In 1896, Bouvert described a syndrome caused by stone impaction in the duodenal bulb leading to gastric outlet obstruction⁷. However, once in the gut, the stone mainly becomes impacted in the terminal ileum causing a small bowel obstruction⁸. A stone greater than 2.5 cm in diameter is often the culprit of bowel obstruction, as those less than 2.5 cm often pass through spontaneously⁸. Uncommonly, a gallstone may become impacted in a Meckel's diverticulum or in the appendix and gallstone ileus may also occur following cholecystectomy⁸. Indeed, gallstone ileus is pretty common in patients over 65 years of age, but the clinical presentation is rather nonspecific and more than one third of patients have no history of biliary symptoms. Thus, a preoperative diagnosis is obtained in only 50 to 60% of patients, and an average period of 3 to 5 days elapse between the first symptoms and the time of hos-

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Correspondence to: Carlo Feo, MD, FACS.UO e Sezione di Clinica Chirurgica, Azienda Ospedaliero-Universitaria di Ferrara, Corso Giovecca 203, 44121 Ferrara, Italy (e-mail: carlo.feo@unife.it)

pital admission^{8,9}. Intestinal obstruction with abdominal pain, vomiting, and constipation is the most common presentation, as seen in 80% of patients reported by Clavien and colleagues². Jaundice is uncommon and is found in less than 15% of patients⁴. Laboratory tests may reveal slight leucocytosis, electrolyte imbalance, and signs of dehydration, while liver function tests are abnormal in about one third of patients. Historically, plain abdominal radiographs were the gold standard in the diagnosis of biliary ileus. Rigler, Borman, and Noble described in 1941 the classic radiologic findings of gallstone ileus, known as Rigler's triad: (1) pneumobilia, (2) direct or indirect visualization of the stone in the gut, and (3) partial or complete bowel obstruction¹⁰. The Rigler's triad, however, occurs in less than 50% of cases on plain abdominal films⁹. It has also been reported that up to half of the patients with gallstone ileus manifest two signs of the classic triad as some gallstone are not radiopaque^{9,11}. The association of ultrasound scanning and plain radiographs of the abdomen has a sensitivity around 74% for definitive diagnosis of gallstone ileus^{8,12,13}. Abdominal computerized tomography may increase sensitivity up to 93% and can identify the cholecystenteric fistula as well as the exact position of the migrated stone¹⁴. In 1890 Courvoisier described a mortality rate for gallstone ileus around 50%; even recently, however, mortality rates as high as 11.5 to 19% have been reported^{2,8}. The reason for this is twofold: 1) most patients are elderly (mean age is approximately 72 years) and therefore present comorbidities, and 2) hospital admission and treatment are often delayed due to the difficult diagnosis⁸.

In this article, an unusual case of biliary ileus is described and the literature on this subject is reviewed and discussed.

Case report

An 83 year-old man presented with abdominal pain and repeated vomiting at the Emergency Department. Past medical history was remarkable for Billroth II gastric resection for peptic ulcer disease, transurethral resection of the prostate (TURP) for benign prostatic hyperplasia, and myelodysplasia with splenomegaly. Interestingly, the patient revealed vomiting a stone two months previously and a three-year history of recurrent colicky abdominal pain; thus, gallstone ileus was suspected. The abdomen was non distended and mildly tender, with no guarding or rebound tenderness, and with hyperactive bowel sounds. The patient was afebrile, white blood cell count was 9'300 mm³, with 80.3% neutrophils, haemoglobin 10.4 g/dl, hematocrit 33.9%, prothrombin time 1.37 INR, blood urea nitrogen (BUN) 104 mg/dl, while the other laboratory tests were unremarkable.

Preoperative plain abdominal radiographs showed dilated loops of proximal small bowel with air-fluid levels.



Fig. 1: The dilated jejunal loop obstructed by the gallstone.



Fig. 2: The 4.5-cm obstructing stone.

An ultrasound scanning of the abdomen demonstrated cholelithiasis, with no signs of cholecystitis or aerobilia. At laparotomy (7 cm) diffuse adhesions were taken down, a single 4.5 cm gallstone was found in the distal jejunum (Fig. 1), and the patient underwent enterolithotomy alone (Fig. 2). The operative time was 50 minutes. The patient's ASA (American Society of Anesthesiologists Classification System) was III. The postoperative course was uneventful and the patient was discharged home five days after the operation, symptom-free, and tolerating an unrestricted diet. The patient died of myelofibrosis four years after the operation; at that time, no episode of bowel obstruction had recurred and he had no sign or symptom of cholangitis, cholecystitis or cancer of the gallbladder.

Discussion

Gallstone ileus should always be suspected in elderly patients presenting with small bowel obstruction, in particular if not previously operated on, and without

inguinal or incisional hernia. Clinically, patients generally present with vague abdominal pain, vomiting, abdominal distension or constipation, and only approximately 50% have a history of gallbladder disease¹⁵. This presentation has been attributed to the so-called “tumbling phenomenon” caused by incomplete small bowel obstruction by the stone⁹. In the presented case, the patient was admitted with small bowel obstruction, he revealed a Billroth II gastric resection and recent vomiting of a stone that suggested passage of gallstones into the gut.

The appropriate treatment in patients affected by biliary ileus is still a matter of debate. The patient with gallstone ileus present two distinct problems: intestinal obstruction and cholecystoenteric fistula. Consequently, two surgical approaches may be followed treating such patients: (1) to address the intestinal obstruction only performing an enterolithotomy (i.e., longitudinal opening proximal to the impacted gallstone, stone extraction, and incision closure) or (2) to address both the obstruction and the closure of the cholecystoenteric fistula.

In the past, the high mortality rates associated to biliary ileus faced the surgeon with the necessity of a rapid procedure to relief the most urgent problem, namely the obstruction⁴. More recently, refinements in the diagnostic imaging may allow earlier diagnosis of biliary ileus and therefore, permit treating a patient in better conditions as compared to the past. Thus, some authors have urged that a definitive operation (i.e., treating the obstruction and the fistula) should be undertaken when treating patients with intestinal obstruction caused by a biliary stone^{3,4,16}. In other words, enterolithotomy with stone extraction, cholecystectomy, and either fistula repair at the same surgical operation (i.e., “one-stage” procedure) or by elective biliary surgery (i.e., “two-stage” procedure). No randomized controlled trial has ever compared these surgical approaches and, most likely, it never will because gallstone ileus is a rare disease.

In the mid-1990's, Reisner and Cohen have reported an extensive review of 1'001 patients with gallstones ileus: 801 (80%) patients treated by enterolithotomy and stone extraction, 113 (11%) by a one-stage surgical procedure, 38 (4%) had other surgical operations, and 49 (5%) received nonoperative treatment⁴. The one-stage procedure was associated with a mortality rate of 16.9% as compared to 11.7% for enterolithotomy alone (P=0.17). Overall, there was a 4.7% recurrence rate, 6% and 5.3% among patients who underwent simple enterolithotomy and one-stage procedure, respectively. Thus, recurrent obstruction occurred even in patients who underwent cholecystectomy because of stone migration from the common bile duct into the bowel. By contrast, 15% of patients undergoing enterolithotomy alone developed biliary complications: specifically, cholecystitis, cholangitis, and gallbladder cancer. This review, however, had several limitations: for instance, none of the studies was randomized and it included series back from 1958 to 1990.

More recently, Doko et al. from Croatia compared intestinal obstruction alone or combined with urgent cholecystectomy and fistula repair in a retrospective series of 30 patients treated for gallstones ileus between 1985 and 2001¹⁷. Operating time was significantly longer for the one-stage procedure (40 minutes vs. 140 minutes, P=0,008). Complications occurred in 27.3% and 61.1% of patients treated for ileus alone and one-stage procedure, respectively (P=0.043). Urgent fistula repair increased the risk of complications (OR 12.1, 95% CI 1.2-121.5). The mortality rate was 9% among patients undergoing enterolithotomy alone versus 10.5% following a one-stage procedure. The author concluded that simple enterolithotomy should be the procedure of choice for patients with gallstone ileus, while the one-stage procedure should be reserved for highly selected patients only. These conclusions substantially confirmed those of a previous retrospective study by Rodríguez-Sanjuán and collaborators from Spain over 25 patients with gallstone ileus¹⁸.

By contrast, Clavien et al from the University of Geneva (Switzerland) reported on 37 patients operated on for gallstone ileus over a 12-year period with a median follow-up of 6.2 years². Specifically, a one-stage procedure was performed in 8 patients, a two-stage procedure in 6, and enterolithotomy in the remaining 23 patients. The three treatment groups were comparable in terms of patient age, comorbidities, and APACHE II score. Interestingly, in 6 patients (16%), more than one stone was present in the bowel. Although operative mortality and morbidity rates did not differ significantly in-between the three groups, later biliary complications prevailed among patients treated by enterolithotomy alone². These results supported the view that a one-stage procedure could be the treatment of choice and alternatively, biliary surgery at a second stage (i.e., two-stage procedure) could be considered when local or surgical conditions argued against a one-stage procedure or if residual stones were present. However, only 10% of patients undergoing enterolithotomy alone and managed expectantly are reoperated on, and the cancer risk seems to increase only for those patients with persistent cholecystoenteric fistula⁴. According to autopsy data, most biliaryenteric fistulas close spontaneously, in the absence of persistent cholelithiasis, after treating the intestinal obstruction and generally, the gallbladder remains shrunken and non-functional⁴.

It is also important to emphasise that multiple stones have been detected into the bowel in 3% to 16% of patients with gallstones ileus; therefore, the entire bowel should be carefully inspected for additional stones¹⁹. Occasionally, the impacted stone causes irreversible damage to the bowel wall and consequently, anastomotic leakage may represent a concerning complication following enterolithotomy. Day and Mark reported a mortality rate as high as 66% following small bowel resection for gallstone obstruction due to anastomotic leakage^{9,20}.

Recently, novel surgical approaches to gallstones ileus have been proposed such as manual delivery of the stone into the cecum followed by colonoscopic extraction and laparoscopic-assisted enterolithotomy¹¹. The problem using laparoscopic techniques is the experience limited to very small number of patients. By laparoscopy, it may be difficult to detect the gallstone in the bowel as well as to safely manipulate the gut, particularly in the presence of dilated loops because of the obstruction. In addition, this practice should not prolong the operative time in critically ill patients. In the presented case, a small laparotomy (7 cm) was performed and a laparoscopic assisted approach would have not offered any additional benefit due to the size of the stone and to the adhesions from previous open surgery.

Conclusions

The evidence on the treatment of gallstone ileus is comprised by small retrospective series of patients recruited over many years. Enterolithotomy alone seems the best treatment option for the majority of patients, while a one-stage procedure may be considered for low-risk patients. Very limited evidence is available regarding interval biliary surgery and so no firm conclusion is possible. A laparoscopic approach may be used in selected cases. It is unlikely that level 1 evidence will ever become available to select the best treatment option in patients with biliary ileus and therefore, such a choice should continue to be based on sound clinical judgement.

Riassunto

L'ileo biliare è una complicanza rara della colelitiasi che costituisce, tuttavia, la causa del 25% delle ostruzioni intestinali senza strangolamento in pazienti oltre 65 anni di età. La morbilità e la mortalità associate all'ileo biliare sono molto alte poichè la diagnosi è spesso tardiva. Il trattamento chirurgico comprende due possibilità terapeutiche: a) enterotomia con estrazione del calcolo ostruente, colecistectomia e riparazione della fistola enterica, eseguita in un solo intervento chirurgico ("one stage procedure" o b) enterotomia con estrazione del calcolo ostruente seguita o meno dall'intervento di colecistectomia in elezione. Quest'ultimo è l'approccio più diffuso e più frequentemente eseguito: infatti, l'enterotomia associata a colecistectomia e riparazione della fistola enterica è il trattamento riservato solo ad alcuni pazienti selezionati.

In questo articolo si descrive un caso di ileo biliare che si è presentato in maniera inusuale ed è stato trattato con enterotomia ed estrazione del calcolo. Il follow up è a lungo termine. È stata inoltre eseguita una revisione della letteratura sull'argomento.

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