

# Bouveret Syndrome: A Curious Simultaneous Presentation of Two Cases: Comparison of Clinical Observations and Surgical Management

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Gallstone ileus is an uncommon occurrence and accounts for about 0.3–0.5% of complications of cholelithiasis in elderly patients. Bouveret syndrome is an uncommon medical condition resulting from the blockage of the duodenal bulb by a stone, which consequently obstructs the outlet of the stomach. Until now, a comparison of two different presentations of Bouveret syndrome has not been published in the literature due to the rarity of this pathology. The curious simultaneous occurrence of the two cases discussed here made it possible for us to compare the different diagnostic and therapeutic pathways. In fact, both cases differ not only in their presenting symptoms, but also in the management adopted by the same surgical team.

**Keywords:** biliary fistula; Bouveret syndrome; cholelithiasis; case report

## Introduction

Gallstone ileus is an uncommon complication of cholelithiasis (0.3–0.5%), mainly affecting elderly patients, and accounts for 1% to 4% of all small bowel obstructions. The increase in life expectancy observed in Western nations is believed to have contributed to a higher occurrence of gallstone ileus, accounting for a quarter of non-strangulated small bowel obstructions among individuals aged 65 and above [1].

Although initially documented by Erasmus Bartholin, a Danish physician, in 1654, it was Courvoisier who published the inaugural series comprising 131 patients in 1890 [2].

Gallstone ileus is not constrained by age, though data from the largest series indicate a mean age of 72 years (with cases ranging from 13 to 97 years) [3].

It occurs more frequently in women than in men, at a ratio of between 4:1 and 16:12. Its pathogenesis involves large gallstones which cause extensive inflammation and adhesion of the gallbladder to the adjacent gastrointestinal tract. The pressure exerted by these stones on the wall causes ischemia, which leads to the erosion of the gallbladder and bowel walls, and the migration of stones into the bowel, creating a cholecystoenteric fistula, with subsequent mechanical bowel obstruction. However, gallstone ileus can also be caused by ulcerating cancers or penetrating ulcers [4].

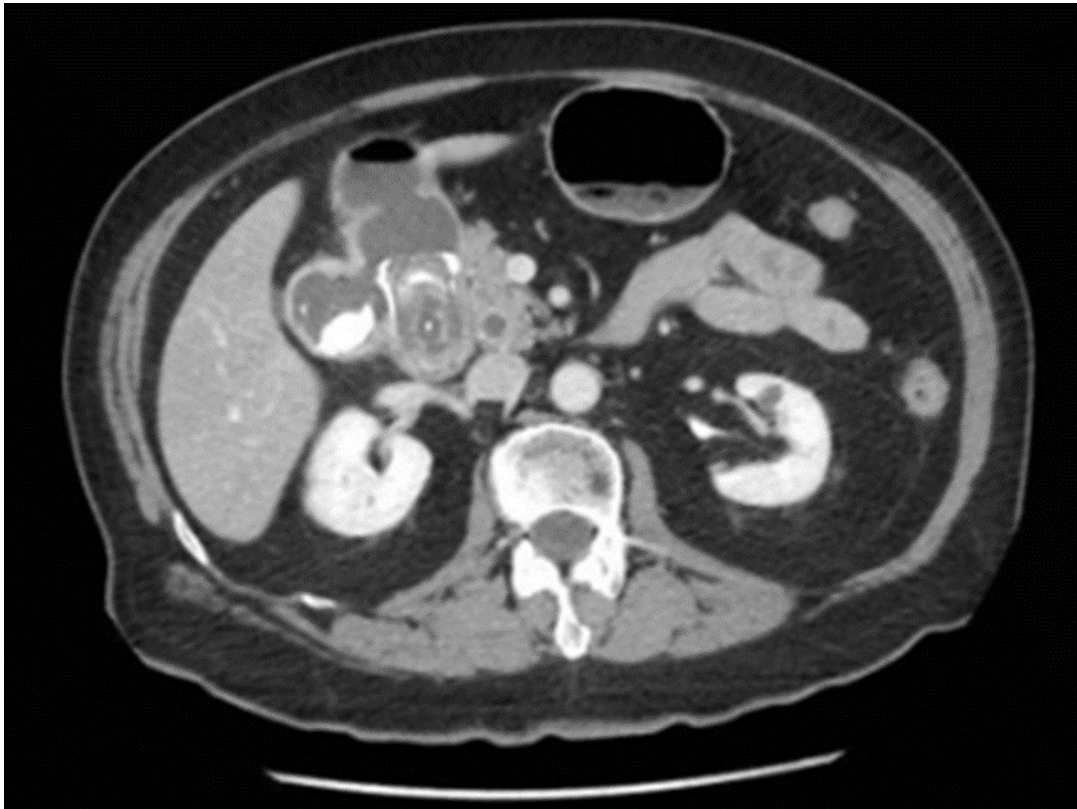
The most common biliary fistula (69%) occurs between the gallbladder and the duodenum (cholecystoduodenal fistula); however, a fistula may also develop between the gallbladder and the ileum (20.3%), the colon (8.5%) or stomach (1.6%) [5].

In 1896, Bouveret described a syndrome caused by stone impaction in the duodenal bulb leading to gastric outlet obstruction. However, once in the gut, the stone can migrate, most commonly to the terminal ileum and become impacted, causing an obstruction [2,6].

A stone greater than 2.5 cm in diameter often causes bowel obstruction, whereas smaller stones may pass freely through the faecal stream. Though rare, a gallstone might become lodged in either Meckel's diverticulum or the appendix. Additionally, gallstone ileus can manifest after a cholecystectomy [7].

In patients aged 65 and older, gallstone ileus is relatively prevalent and typically presents with vague symptoms. Surprisingly, over a third of these patients have no prior history of biliary issues, resulting in a preoperative diagnosis occurring in only about 50 to 60% of cases, typically after 3 to 5 days following symptom onset and hospital admission. Laboratory tests might indicate mild leukocytosis, electrolyte imbalances, and signs of dehydration, with around one-third of patients displaying abnormal liver function test results. Traditionally, plain abdominal radiographs were considered the diagnostic standard for biliary ileus. In 1941, Rigler *et al.* [8] delineated the classic radiological features of gallstone ileus, termed Rigler's triad: pneumobilia, direct or indirect visualization of the stone within the gut, and partial or complete bowel obstruction. However,

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**Fig. 1. Abdominal computed tomography (CT) scan of the first patient.** The gallbladder is distended, containing multiple lithiasic formations, the largest approximately 22 mm, with diffuse thickened and edematous walls.

Rigler's triad is observed in fewer than 50% of gallstone ileus cases [5].

Up to half of the patients with gallstone ileus manifest only two features of the classic triad, because some gallstones are not radiopaque [5,7,9].

To date, a comparison of two different presentations of Bouveret syndrome has not been published in the literature due to the rarity of this pathology. The simultaneous finding of these two cases at the same time period at our institution has offered us the opportunity to compare the diagnostic and therapeutic pathways, which differ not only in onset symptoms but also in the diagnostic and surgical strategies adopted by the same surgical team.

The aim of this article is to present two cases of patients admitted in our hospital in the same period, with diagnoses of cholelithiasis and cholecystoduodenal fistula with migration of gallstone into the duodenum, and to assess the diagnostic and therapeutic strategies and compare the outcome of both cases.

### Case 1

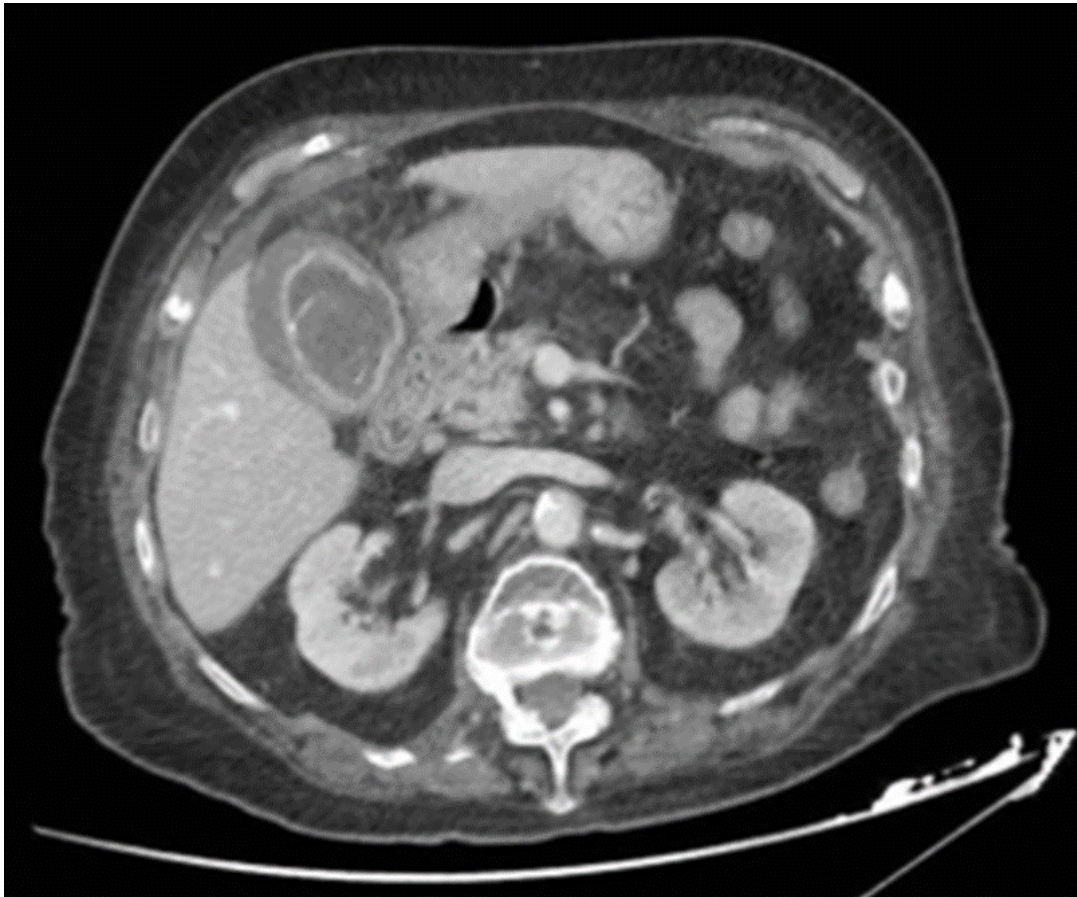
A 75-year-old woman with a past medical history of type II diabetes and arterial hypertension was admitted for epigastric pain, nausea, and vomiting. The clinical examination showed sub-jaundice, and a positive Murphy sign. There was no sign of peritoneal involvement. Laboratory tests

showed a normal white blood cell count; total serum bilirubin level was 2.08 mg/dL with a direct component of 1.29 mg/dL; C-reactive protein (CRP) was 1.29 mg/dL; and Procalcitonin was 0.36 ng/mL. An abdominal ultrasound scan showed cholecystitis with cholelithiasis, intrahepatic biliary tract dilatation, and aerobilia. A contrast-enhanced abdominal computed tomography (CT) scan was performed and showed a 21 mm fistula between the gallbladder and duodenum, and a large gallstone of more than 44 mm in the duodenum. The stomach was distended (Fig. 1). The CT confirmed aerobilia, especially involving the left biliary tract and the gallbladder. An open cholecystectomy and a duodenal wall excision, which included the cholecystoduodenal fistula, were performed.

Duodenal wall repair was performed with continuous suture using PDS 4.0 in a triple layer. Cyan acrylate (Glubran) glue was also applied along the suture line.

In the postoperative period, the patient was admitted to the intensive care unit for 5 days. Due to persistent nausea, the nasogastric tube was maintained for the first 8 days, and the patient resumed feeding on the 10th day after the surgery. No further complications occurred during the hospitalization, and the patient was discharged after 17 days, following a physiotherapy program.

Two weeks later, the patient was readmitted due to abdominal pain without fever. Suspecting a possible abdominal infection, a contrast-enhanced CT scan identified an ab-



**Fig. 2. Abdominal CT, second patient.** The gallbladder has heterogeneous hyper-dense contents with thickened walls, fluid collection, and an endoluminal lithiasis formation of about 30 mm, poorly calcified.

cess in the gallbladder fossa. The patient received a broad-spectrum intravenous antibiotic therapy with piperacillin-tazobactam at a dose of 18 grams per day.

A double-contrast intestinal X-ray with gastrografin did not detect intestinal dehiscences, and esophago-gastroduodeno gastroscopy (EGDS) revealed the presence of a duodenal ulcer and a probable small fistula of the intestinal wall. This condition was confirmed after a subsequent abdominal CT scan. Consequently, an echo-guided drainage of the abdominal collection and the placement of a Padlock-type clip via endoscopy were performed. Subsequent ultrasound (US) checks documented the resolution of the abdominal abscess, and the patient completed the antibiotic therapy, being discharged 50 days after the initial hospitalization.

## Case 2

An eighty-one-year-old woman was admitted with a past medical history of arterial hypertension, previous brain stroke, chronic cerebral vascular disease, poly-allergy, and previous surgery for vaginal prolapse. The patient presented to the hospital with fever and diffuse abdominal pain. During the clinical examination, positive Murphy and Blumberg signs were observed. Laboratory tests showed el-

evated white blood cell count and CRP (21.9 mg/dL), and a creatinine level of 1.85 mg/dL. The abdominal US scan revealed cholecystitis and multiple gallstones (the largest one measured 25 mm in diameter). For a comprehensive diagnostic evaluation, an abdominal CT scan with contrast was performed.

The CT revealed the presence of gallstones in the gallbladder and an 8 mm stone within the common bile duct. This situation necessitated an Endoscopic Retrograde Cholangiopancreatography (ERCP), but the bile duct could not be cleared due to the unsuccessful cannulation of the Vater's papilla.

Consequently, the patient underwent laparotomy and cholecystectomy, during which a single stone of approximately 30 mm was extracted (Fig. 2). The diagnosis of Bouveret syndrome with a cholecystoduodenal fistula was confirmed during the procedure. An intraoperative cholangiogram was performed, and a second attempt at ERCP failed to clear the main bile duct. To complete the procedure, a Kehr tube was inserted, and the duodenal fistula was interrupted and sutured using a linear mechanical stapler Ecelon Flex 45 mm (1 blue load), reinforced with manual stitches using PDS 4-0.

The patient was kept in the intensive care unit for 3 days before returning to our department. On the 7th postoperative day, an ultrasound revealed asymptomatic residual cholelithiasis. Ten days after the procedure, a repeat ERCP with sphincterotomy allowed the extraction of a residual stone, measuring 7 mm, from the bile duct. During the hospital stay, a rehabilitation program was necessary for a gradual recovery of physical functions. Forty-two days after the procedure, the Kehr tube was removed.

Despite recovery from the abdominal infection, a rapid clinical deterioration occurred due to heart failure and acute renal insufficiency with dyspnea, leading to the patient's demise.

## Discussion

Gallstone ileus should always be taken into consideration in elderly patients with past medical history of biliary disease, who present with small bowel obstruction, especially when accompanied by vague abdominal pain, vomiting, abdominal distension, or constipation [10].

The optimal treatment for gallstone ileus remains a subject of debate. Patients with biliary ileus present two distinct challenges: intestinal obstruction and cholecysto-enteric fistula. Therefore, the surgeon may follow one of these two different approaches to treat this condition:

1 - to solve the problem of intestinal obstruction by performing an enterolithotomy (A longitudinal incision on the impacted bowel, close to the impacting gallstone, extraction of the stone, and closure of enterotomy).

2 - to treat both the obstruction and close the cholecysto-enteric fistula [10].

Historically, the elevated mortality rates linked with biliary ileus prompted surgeons to prioritize swift intervention to address the most pressing issue: the obstruction [5,10].

In recent years, improvements in diagnostic imaging have allowed for quick diagnosis of small intestinal occlusion caused by gallstones and have permitted treatment of these patients in better conditions. Hence, some authors have suggested that definitive operations (that is, combined enterolithotomy with stone extraction, cholecystectomy, and fistula repair) should be done at the same time when treating patients with intestinal obstruction caused by a biliary stone [4, 6, 10]. Despite the availability of new surgical technologies and techniques, patient factors, such as co-morbidities and medications, can also influence the outcome of treatment.

We have reported two different therapeutic approaches to the same pathology: the first, a one-stage treatment involving cholecystectomy and repair of the duodenal fistula; the second, gallbladder drainage followed by endoscopic clearance of the bile duct. Furthermore, in the first case, duodenal repair was performed using three rows of manual sutures with the addition of fibrin glue, whereas in the second case, mechanical suturing was employed and manually reinforced. In both cases, no leakage occurred.

Based on our experience, we suspect that the adverse pre-operative clinical conditions of the oldest patient, including fever, and high levels of inflammatory markers, and the chronic vascular disease, contributed to more postoperative complications. Interestingly, in our first case, the patient, despite being diabetic, did not exhibit fever, pain, or neutrophilic leukocytosis. However, she later developed an abdominal infection. In contrast, the comorbidities of the second patient gradually led to multiorgan failure.

According to the study conducted by Reisner *et al.* [6], 15% of patients who solely underwent enterolithotomy experienced subsequent biliary complications such as cholecystitis, cholangitis, and gallbladder cancer. Interestingly, the mortality rate among patients undergoing enterolithotomy alone (9%) was comparable to that following a one-stage procedure (10.5%). As a result, the authors advocated for simple enterolithotomy as the preferred procedure for patients with gallstone ileus and suggested that the one-stage procedure should be reserved for highly selected patients only [11,12,13].

Doko *et al.* [14] conducted a retrospective analysis comparing the treatment of intestinal obstruction alone versus combined urgent cholecystectomy and fistula repair in a series of 30 patients treated for gallstone ileus between 1985 and 2001 [8,15,16].

The one-stage procedure exhibited significantly longer operating times compared to treating intestinal obstruction alone (40 minutes vs. 140 minutes,  $p = 0.008$ ). Complications were observed in 27.3% and 61.1% of patients undergoing treatment for ileus alone versus the one-stage procedure, respectively ( $p = 0.043$ ). Urgent fistula repair was associated with an increased risk of complications (OR 12.1, 95% CI 1.2–121.5). The mortality rates were comparable between patients undergoing enterolithotomy alone (9%) and those undergoing the one-stage procedure (10.5%). Consequently, the authors recommended simple enterolithotomy as the preferred approach for patients with gallstone ileus, reserving the one-stage procedure for highly selected cases. Moreover, it is noteworthy that multiple stones have been discovered in the bowel in 3% to 16% of patients with gallstone ileus, underscoring the importance of thorough inspection of the entire bowel to exclude the presence of additional stones. In recent times, some studies have suggested laparoscopic-assisted enterolithotomy as a potential approach [16,17].

The problem with using laparoscopic techniques is the limited experience with a very small number of patients.

During laparoscopy, safe manipulation of the gut in search of obstructing gallstone may be challenging, particularly in the presence of dilated loops due to the obstruction [17,18,19].

Surgical or endoscopic treatments should be tailored to suit the patient's medical conditions and clinical presentation. A laparoscopic approach has been shown to be successful in removing stones up to 5 cm [20].



## Conclusions

Enterolithotomy alone appears to be the most suitable treatment option for the majority of patients with gallstone ileus, whereas a one-stage procedure might be considered for low-risk individuals. However, the available evidence for the optimal therapy remains limited, precluding firm conclusions. It is improbable that robust evidence will ever emerge to definitively determine the best treatment option for biliary ileus. Thus, such decisions should continue to be guided by sound clinical judgment. In selected cases, a laparoscopic approach can be employed by experienced surgeons.

## Availability of Data and Materials

The datasets used or analyzed during the current study are available from the corresponding author on reasonable request.

## Author Contributions

Conceptualization: NF, IA; methodology: NF, FV, IA; validation: NF, DR, IA; formal analysis: NF, DR; investigation: NF, FV, IA; resources: NF, DR; data curation: NF, DR; writing—original draft preparation: NF, FV; writing—review and editing: NF, DR, IA; visualization: NF, DR; supervision: NF, IA. All authors have read and agreed to the published version of the manuscript. All authors have participated sufficiently in the work and agreed to be accountable for all aspects of the work.

## Ethics Approval and Consent to Participate

The case reports was exempted from ethical approval by Local Health Unit of Ferrara based on art 3 D.M. 30 January 2023 delibera della Regione Emilia-Romagna num.1029/19. 06. 2023 seduta num. 27. Patients written consent was obtained for this publication. The study follows the Declaration of Helsinki.

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## Conflict of Interest

The authors declare no conflict of interest.

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