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# Gallstone ileus: analysis of eight cases and review of the literature

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**Rezumat**

***Ileusul biliar: analiza a opt cazuri şi recenzia literaturii***

*Scop:* propunem o analiză a cazurilor de ileus biliar operate în clinică, prilejuită de un caz de sindrom Bouveret.
*Metoda:* studiu retrospectiv al cazurilor de ileus biliar operate în clinica noastră în ultimii 26 de ani, urmărind elementele diagnostice, intervalul scurs până la operaţie, tipul intervenţiei şi evoluţia post-operatorie.
*Rezultate:* în această perioadă s-au operat 9143 litiaze biliare descoperindu-se 27 fistule biliodigestive din care 8 s-au complicat cu ileus biliar. Ileusul a survenit exclusiv la femei vârstnice cu comorbiditate importantă. Diagnosticul sugerat de semnele clinice ale ocluziei acute sau subacute, a fost susţinut de investigaţii imagistice a căror valoare diagnostică a fost diferită. Intervalul scurs de la internare până la operaţie a fost de 2,6 zile. Atitudinea chirurgicală a oscilat între simpla enterolitotomie şi asocierea acesteia cu desfinţarea fistulei biliodigestive. Evoluţia a fost favorabilă în majoritatea cazurilor cu o excepţie (1 deces).
*Concluzii:* ileusul biliar este o afecţiune rară, survenită la vârstnici cu comorbiditate importantă. Alegerea procedeului chirurgical depinde de impactul sidromului ocluziv şi de comorbiditatea asociată, tipul intervenţiei neinfluenţând semnificativ morbiditatea şi mortalitatea post-operatorie.

**Cuvinte cheie:** fistula colecisto-duodenală, ileus biliar, sindrom Bouveret

**Abstract**

*Objective:* We hereby analyzed a series of gallstone ileus cases operated on in our department starting from a Bouveret syndrome case.
*Method:* Retrospective analysis of all gallstone ileus cases who underwent surgery in our department during the last 26 years. We took into consideration diagnostic elements, time from admission to surgery, type of surgery and post-operative outcome.
*Results:* During this period 9,143 gallstones were deferred to surgery; 27 biliary-digestive fistulae were discovered during surgery; gallstone ileus complicated fistula in 8 patients. Gallstone ileus was exclusively present in elderly women with associated comorbidities. Diagnosis was suggested by clinical features of acute or incomplete intestinal obstruction; it was sustained by imagistic studies with different degrees of relevance. The average time from admission to surgery was 2.6 days. Surgical approach varied from simple enterolithotomy to additional fistula repair. The outcome was uneventful in most of the cases with only one exception.
*Conclusions:* gallstone ileus is a rare condition, occurring in elders with important comorbidities. The choice for surgical procedure depends on the obstructive syndrome's gravity and associated comorbidities; the type of intervention does not significantly influence post-operative morbidity and mortality rates.

**Key words:** gallblader-duodenal fistula, gallstone ileus, Bouveret syndrome

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## Introduction

Starting from one case of duodenal obstruction by a migrated gallstone through a gallbladder-duodenal fistula, we analyzed all patients with gallstone ileus in our surgical department and reviewed the literature. Gallstone ileus, first

described by Bartholin in 1654, is a major but rare complication of billiary-digestive fistulae due to gallstone migration in the digestive tract causing its obstruction(1). In association with mechanical and septic factors gallstones may complicate with billiary-digestive fistulae in 0.15 – 5 % of cases (2,3,4,5) *(Fig. 1)*. Alongside with anaemia and angiocolitis, gallstone ileus is one of the billiary-digestive fistulae's complications – in 14% of cases(1,6). Generally, gallstone ileus represents a rare cause of intestinal obstruction (1-4%), but in elderly patients (over 65 years) can cause up to 25% of unstrangulated intestinal obstructions (2,3,7).

## Method

Analysis was performed retrospectively on 8 patients with gallstone ileus who underwent surgery in our surgical department over the past 27 years. Evaluation was focused on symptoms, comorbidities and studies which confirmed the preoperative diagnosis, the time between admission to surgery, intraoperative findings, surgical procedure, post operative outcome and mortality rate. We also reviewed medical literature.

## Results

62,126 surgical procedures were performed in our department between 1980-2007, out of which 9,143 were for gallstones. 27 billiary-digestive fistulae were discovered during surgery, 8 of these complicated with gallstone ileus.

717 patients underwent surgery for intestinal obstruction in the same period of time; those 8 patients represent 1.15% of all intestinal obstructions.

The 8 cases of gallstone ileus occurred only in females aged between 50 – 87 years, with an average age of 71.5.

The history of gallstones was present only in 3 patients.

As comorbidities, all patients had diabetes mellitus associated with peripheral neuropathy and microangiopathy. Moreover, the majority of them had obesity, dislipidemia, blood hypertension and ischemic heart disease. Two patients were recorded with chronic kidney failure and varicose veins.

Symptoms of intestinal obstruction occurred in all patients; they were acute in 4 patients (i.e. acute abdominal pain, vomiting – intestinal fluid-like) and mild for the rest (i.e. intermittent episodes of colicative abdominal pain, bilious vomiting with free interval of several days).

The time between patient's admission and surgery was 2.6 days.

In our series plain radiography showed elements of Riegler's triad (8). Dilated small-bowel loops with air fluid levels suggestive of intestinal obstruction were seen in all patients except for the case with duodenal obstruction. Pneumobilia as pathognomonic sign occurred only in 3 patients *(Fig. 2)*. Contrast X-Ray showed gallstone only in patient with Bouveret syndrome *(Fig. 3)*.

Only in the patient with Bouveret syndrome endoscopy identified duodenal obstructive gallstone but the extraction attempt was unsuccessful.

Abdominal ultrasonography revealed dilated intestinal loops and pneumobilia in 2 cases.

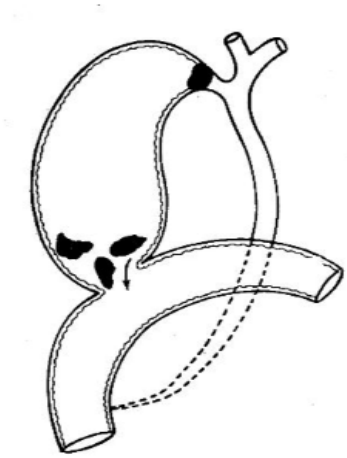

***Figure 1.*** *The pathophysiology of billiary-digestive fistulae (1)*

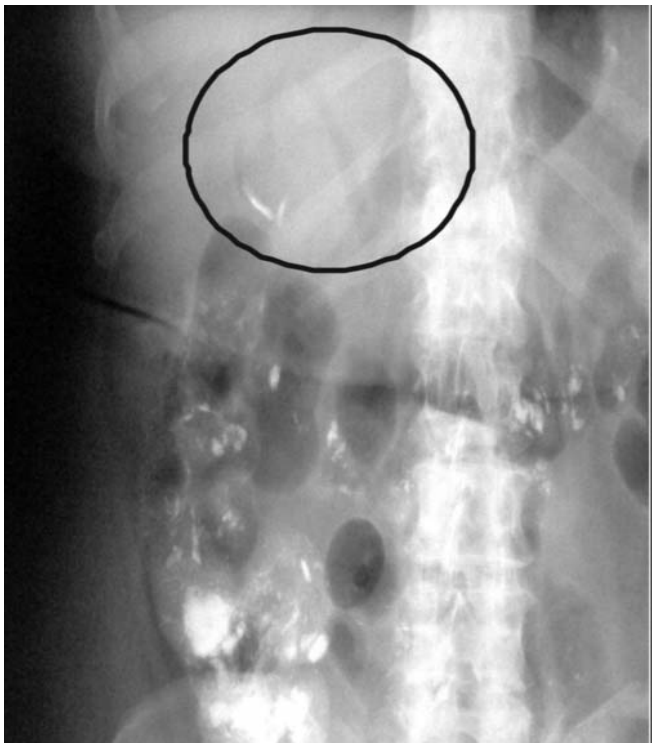

***Figure 2.*** *Supine X-Ray showing pneumobilia*

Bilio-enteric fistulae were directly identified during surgery in 3 patients. For the other 5 where the only surgical approach was enterolithotomy pseudo-tumoural mass like was founded in the right upper quadrant at the time of surgery which indirectly led to the assumption of fistula presence.

The locations of gallstone obstruction were the duodenum (1 patient), jejunum (3 patients), terminal ileum and ileo-cecal valve (4 patients). Multiple gallstones were identified only in 1 patient, whereas the other 7 presented a single gallstone

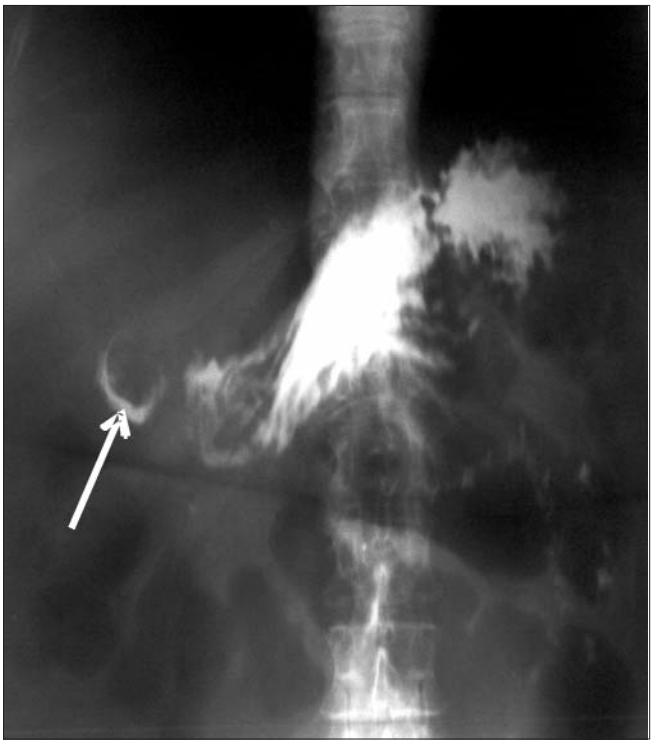

***Figure 3.*** *Oral contrast X-Ray suggesting a duodenal gallstone*

obstruction each. The size of the gallstones varied from 2.5 to 8 cm.

Surgical approach addressed only intestinal obstruction in 6 of the patients and consisted in enterolithotomy in 4 of them. In one patient, unexpected intestinal lesions required minimal enterectomy. In the 6th patient (duodenal obstruction), after an unsuccessful attempt of endoscopic removal, the fragmented gallstone was surgically extracted through an antral gastric incision. Another piece of the gallstone was extracted through a gallbladder incision followed by suture of the gallbladder wall. The post-operative endoscopy showed a large cholecysto-duodenal fistula with a pseudo diverticular aspect of the gallbladder, but without remnant fragments of stones.

The one-stage procedure composed of enterolithotomy, cholecystectomy and repair of fistula was performed in 2 out of 8 patients. In both patients, the extraction of gallstones through enterolithotomy was followed by partial cholecystectomy in order to avoid dissection of the inflammatory modified cystic region. In the first patient the repair of the cholecystoduodenal fistula was performed by transversal duodenoraphy (about 0.7 cm diameter). In the second case, because the duodenal orifice was large enough (about 2/2.5 cm diameter) it was sutured around a Pezzer catheter (exteriorized in the right upper quadrant) after antrectomy, gastric-jejunal anastomosis and vagotomy as a duodenal diversion.

Post-operative outcome was uneventful in 5 patients (55%). One of the enterolithotomy patients developed a postoperative fistula in the 8th post-operative day. Enterectomy followed by jejunojejunal anastomosis was performed but unfortunately the patient died 2 days later of septic shock. A stroke occurred in one patient and another developed an infection at the site of the incision. The patients operated on only with enterolithotomy never come back for surgical treatment of the remnant fistula or for another episode of gallstone ileus. Only one patient was readmitted a year later for a new episode of intestinal obstruction. During laparotomy a pseudo-tumoural mass in the right quadrant was identified. By antral gastrotomy a complete duodenal stenosis was found and a gastro-jejunal anastomosis was performed, with good postoperative outcome.

The average hospitalization period was 14.8 days and the mortality rate was 12,5% (1 patient).

## Discussions

Gallstone is a common disease with prevalence in 10% of the adult population in the United States and Western Europe (9). The most common complications of gallstone disease include acute cholecystitis, acute pancreatitis, choledocholithiasis with or without cholangitis, and a gangrenous gallbladder. Other uncommon complications include Mirizzi syndrome, cholecystocholedochal fistula, and gallstone ileus (10, 11).

Gallstone ileus affects mainly elderly patients with important comorbidities, dominated by diabetes mellitus and atherosclerosis in our series.

Although the ethiopathogeny of the condition consists of mechanical factors (e.g. cystic or common billiar duct obstruction, erosion of gallbladder's walls) and/or septic factors (e.g. infection of gallbladder or digestive tract) half of our patients had no history or clinical features of gallbladder disorder. The gallstone enters the GI tract through a fistula between gallbladder and duodenum, stomach or colon. It can impact anywhere in the GI tract and its size should be at least 2 cm to 2.5 cm in diameter to cause obstruction (12). Although the most common locations of occurrence for gallstone are the terminal ileum and the ileocecal valve because of its anatomical small diameter, they were also found less common in the jejunum and the stomach, while the duodenum and colon are the rare locations for their occurrence.

The clinical findings suggest from the beginning a complete intestinal obstruction. Other times clinical features advocate episodes of incomplete intestinal obstruction following intermittent gallstone migration at different levels of the digestive tract, eventually leading to complete blockage (2).

Laboratory studies may show an obstructive pattern with electrolytes imbalances and leucocitosis.

The preoperative diagnosis is exclusively based on imaging studies with different but complementary value (13). Often the diagnosis is only made at laparotomy.

The classic Rigler's triad of radiography includes mechanical bowel obstruction, pneumobilia, and an ectopic gallstone within bowel lumen (14). Plain abdominal films usually show non-specific findings because only 10% of gallstones are sufficiently calcified to be visualized radiographically.

In our series the abdominal X-Rays identified pneumobilia (a pathognomonic sign) in 3 patients (37%) and only in the patient with Bouveret syndrome showed the gallstone.

In the case of the Bouveret syndrome, the endoscopy may show duodenal gallstones obstruction and may treat the obstruction successfully. During endoscopy the fistula may be seen, as well as the presence of remnant gallstones in the gallbladder.

Abdominal echography is useful in confirming the presence of cholelithiasis; it may identify fistula, if present (15). Abdominal US confirmed the presence of pneumobillia in 2 patients on our series.

Abdominal CT becomes the more important modality in diagnosing gallstone ileus because of its better resolution. CT scan may expose the migrated gallstones (92%) and it is superior to sonography or X-Rays in revealing pneumobilia (13). CT scan was not used in our lot.

Lassandro et al compared the clinical value of plain abdominal film, abdominal US and abdominal CT in diagnosing 27 cases of gallstone ileus, and found that the Rigler's triad presents 14.81% in plain abdominal film, 11.11% in abdominal US, and 77.78% in abdominal CT, respectively. (13)

Additionally, Yu et al studied the value of abdominal CT in the diagnosis and management of gallstone ileus and concluded that the abdominal CT offers crucial evidence not only for the diagnosis of gallstone ileus but also for decision making in management strategy (16).

Rarely, laparoscopy is used to diagnose this disease (17).

Delayed diagnosis can be caused by inconclusive imaging (absence of pneumobilia – 50-60% cases), absence of CT scan and intermittent or incomplete onset of intestinal obstruction (2,6).

Gallstone ileus usually requires emergent surgery to relieve intestinal obstruction. There is no uniform surgical procedure for this disease because of its low incidence. Surgical options (enterolithotomy with or without fistula treatment) are linked to the impact of acute intestinal obstruction and associated comorbidities of the patients. Bowel resection is indicated only when there is intestinal perforation or ischemia (18).

The endoscopic gallstone removal in duodenal obstruction may fail in the absence of ultrasound gallstones breaking devices and call for surgery treatment (19, 20 and 21).

Although a simple procedure, enterolitothomy may lead to lethal complications (one of our patients died of septic shock following post-operative fistula).

In remnant fistula intestinal obstruction may reoccur by migration of new gallstones; it may also complicate with: chronic inflammatory duodenal stenosis, angiocolitis, anaemia, and gallbladder's carcinoma (2). The postoperative recurrence rate of gallstone ileus is 4.7% and only 10% of patients require secondary biliary surgery for recurrent biliary symptoms (7, 23).

Fistula removal in difficult dissection conditions can call for an incomplete cholecystectomy with reduced rate of biliary duct injury. A large duodenal fistula (1.5 – 2 cm) can require a duodenal diversion through a gastric-jejunal anastomosis.

The prognosis of gallstone ileus is usually poor and worsens with age. Though morbidity and mortality decreased over time from 50% to 18% (12.5% for our group), they are still high largely due to delayed diagnosis and concomitant conditions such as cardio-respiratory disease, obesity and diabetes mellitus (2, 6 and 24). The decrease of mortality and morbidity is foremost due to per-operative therapy rather than surgical attitude, which have remained constant (2, 6)

In conclusion, gallstone ileus is a rare cause of intestinal obstruction. It must be considered in intestinal obstruction patients with a history of gallstone, especially in elderly women. Surgical treatment is emergent when the radiological finding is highly or even suspicious or confirmed.

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