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An Interesting Presentation of Bouveret Syndrome: Tumbling Phenomenon and Possible Genetic Component

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Background	Gallstone ileus, a rare complication of cholelithiasis, occurs when a gallstone migrates from the gallbladder and causes obstruction within the small intestine. This typically happens due to a biliary-enteric fistula. In some cases, the gallstone may lodge in the stomach or duodenum, causing gastric outlet obstruction. This specific scenario is known as Bouveret syndrome.
Summary	This report describes an unusual case of gallstone ileus in a 74-year-old woman with no prior gallbladder issues but a positive family history for the condition. The patient presented to the emergency department with symptoms suggestive of small bowel obstruction, including abdominal pain, nausea, and vomiting. A CT scan revealed a 2 cm radiopaque object within the duodenum, consistent with a gallstone, along with moderate stomach dilation. Endoscopy confirmed the presence of the gallstone in the duodenum. However, during the subsequent enterolithotomy, the gallstone was surprisingly found impacted within the jejunum. Ultimately, the patient tolerated the surgery well and recovered without complications.
Conclusion	This case presentation holds significance for several reasons. Both gallstone ileus and Bouveret syndrome are rare complications associated with gallstones. Additionally, this case highlights the "tumbling phenomenon" and demonstrates how gallstone ileus can initially present as Bouveret syndrome and subsequently evolve into a jejunal impaction. Lastly, this case raises the possibility of a genetic predisposition to gallstone ileus, considering the patient's family history.
Key Words	gallstone ileus; Bouveret syndrome; intestinal obstruction; bowel obstruction; enterolithotomy

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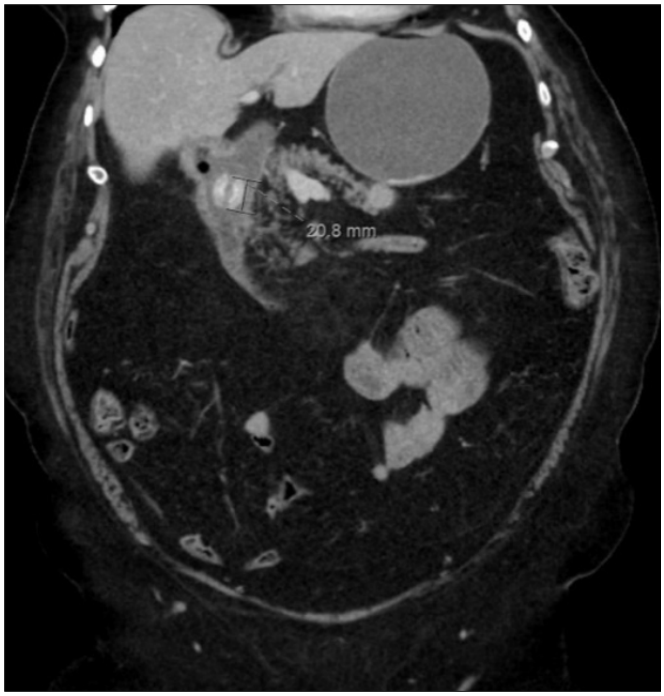
Case Description

A 74-year-old white female presented to the emergency department with a 36-hour history of nausea, vomiting, and severe epigastric abdominal pain. She had no significant medical history but reported gradually worsening burning epigastric pain over the preceding two weeks. Her primary care physician trialed her on 40 mg of omeprazole for the aforementioned symptoms, but it provided no relief. Liver function tests were mildly abnormal (bilirubin 1.5 mg/dL, alkaline phosphatase 376 U/L, alanine aminotransferase 88 U/L, aspartate aminotransferase 38 U/L) and an elevated white blood cell count (16.8 × 10^9/L).

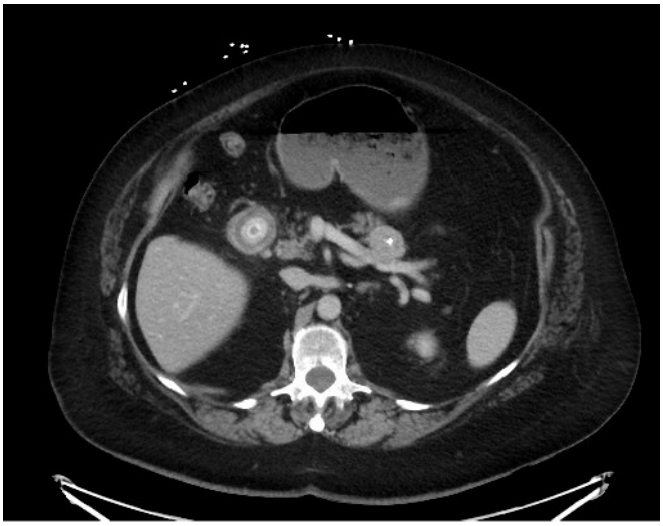
A computed tomography (CT) scan demonstrated a 2 cm rounded, radiopaque structure within the second portion of the duodenum, obstructing the outflow of stomach contents. The stomach appeared moderately dilated. The gallbladder was not clearly visualized, suggesting inflammation with erosion into the duodenum, stomach, or hepatic flexure. The majority of gallstone impaction sites are in the ileum related to the smaller diameter of the distal small bowel. Mild intrahepatic biliary ductal dilation, most significant in the left hepatic lobe, was observed (Figures 1 and 2).

Figure 1. Coronal CT Imaging of Gallstone Ileus. Published with Permission



Coronal CT image demonstrating a gallstone within a small bowel loop in the right upper quadrant, with no evidence of distal small bowel dilation.

Figure 2. Axial CT Imaging of Gallstone Ileus. Published with Permission



Axial CT image demonstrating a gallstone within a small bowel loop in the right upper quadrant.

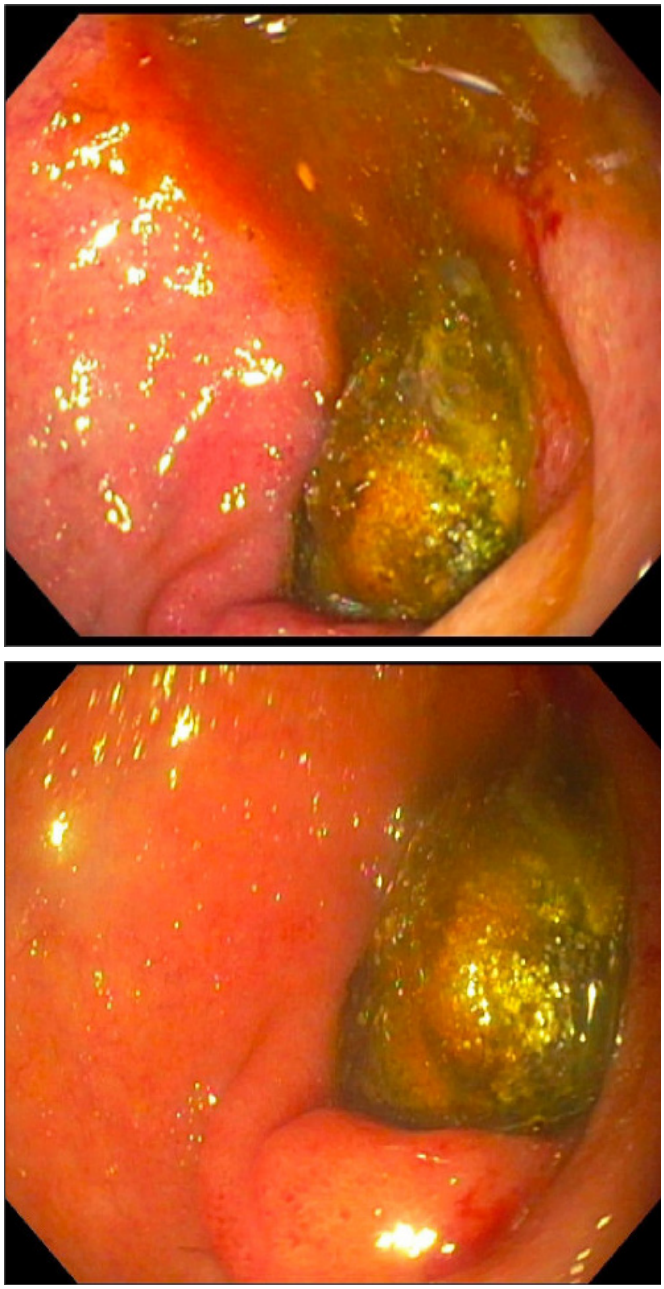
Given the absence of prior gallbladder resection and the constellation of findings, concern for Bouveret syndrome—a rare complication where a gallstone migrates from the gallbladder and becomes lodged in the duodenum, causing obstruction—was raised. The patient was admitted for further management. Initial interventions included fluid resuscitation, nil per os (NPO), placement of a nasogastric tube for decompression, and appropriate analgesia and anti-nausea medications. A Foley catheter was placed to monitor urine output.

The gastroenterology team attempted esophagogastroduodenoscopy (EGD) to visualize the obstruction. However, the cholecystoduodenal fistula could not be visualized due to the size and immobility of the stone. Attempts to dislodge the stone or place a Roth net were unsuccessful.

Following consultation with general surgery for gastric outlet obstruction, possibly due to Bouveret syndrome, the patient consented to an exploratory laparotomy with potential bowel resection and open cholecystectomy. Surgical intervention proceeded within 24 hours. While no stone was initially palpable in the proximal small bowel, further exploration revealed a single palpable stone lodged within the jejunum. A 2 cm longitudinal enterotomy was created using electrocautery to access the stone, which was then successfully delivered from the bowel lumen (Figure 3). The enterotomy was closed in an interrupted fashion with 3-0 silk sutures. Additionally, a falciform flap was fashioned and positioned in the right upper quadrant near

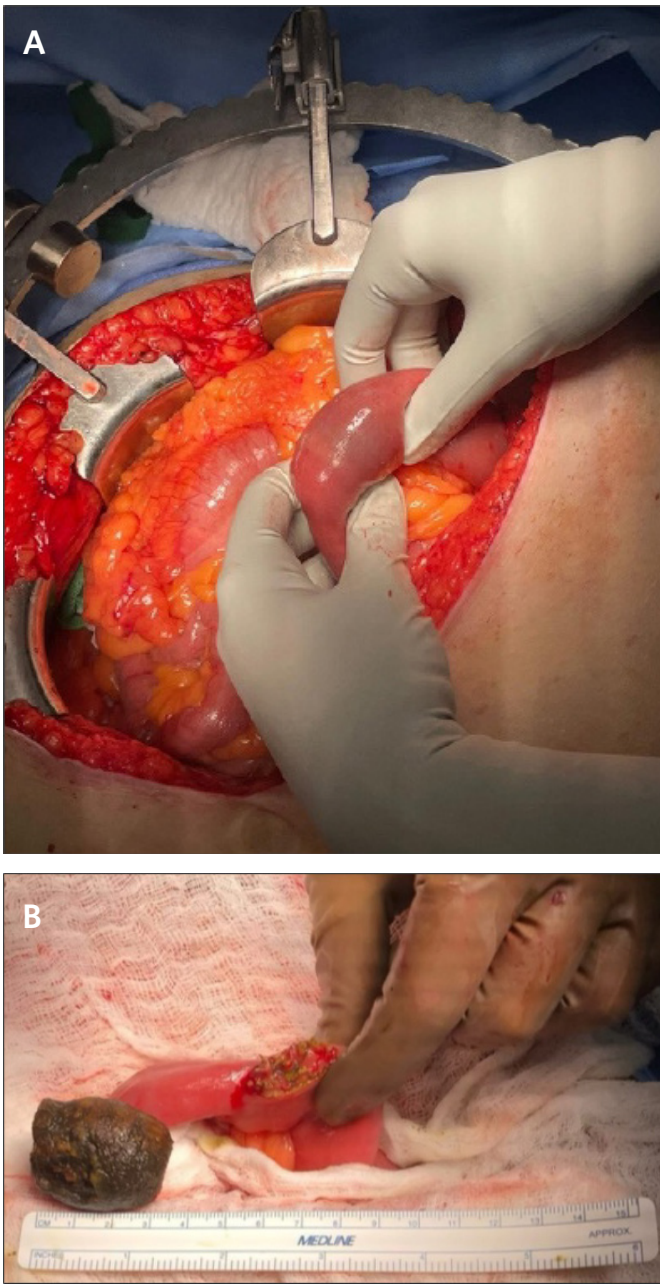
a visualized cholecystoduodenal fistula. The patient tolerated the procedure well, was extubated in the operating room, and taken to the postanesthesia care unit (PACU) in excellent condition. Her postoperative recovery was uneventful, and she was discharged home four days after the procedure.

Figure 3. Endoscopic Identification of Duodenal Gallstone. Published with Permission



Gallstone impacted within the second portion of the duodenum, as identified via upper endoscopy.

Figure 4. Gallstone in the Jejunum. Published with Permission



A) *Endoscopic visualization of a gallstone within the proximal jejunum.* ***B)*** *Surgical retrieval of the gallstone via enterolithotomy.*

Discussion

Gallstone ileus, though encountered very infrequently, represents a potential late complication of gallstones and a rare cause of intestinal obstruction, accounting for only 1% of such cases.[1] Typically affecting elderly and female patients, gallstone ileus arises when a gallstone migrates

from the gallbladder into the small intestine through a biliary-enteric fistula, most commonly formed between the gallbladder and duodenum.[2] A high index of suspicion for gallstone ileus is crucial in patients with a known history of gallstones who present with bowel obstruction.

The mainstay of treatment involves mechanically removing the impacted gallstone(s) either through an open or laparoscopic approach.[3] Enterolithotomy is the preferred surgical method due to its lower complication incidence.[4] While abdominal X-ray and ultrasound are often the initial imaging studies for acute abdominal pain, they are not sensitive for diagnosing gallstone ileus. For definitive diagnosis and to identify alternative etiologies, CT is the imaging modality of choice for this disease.[5] An even rarer complication of gallstone disease is Bouveret syndrome, where the gallstone lodges in the stomach or duodenum, causing gastric outlet obstruction. This presentation constitutes only 1-3% of gallstone ileus cases.[6,7]

The development of gallstone ileus typically follows a pattern of recurrent cholecystitis episodes. Chronic inflammation of the gallbladder can lead to fistula formation between the gallbladder and the gastrointestinal tract. This newly created passage allows a gallstone to migrate into the gastrointestinal (GI) tract, resulting in mechanical bowel obstruction. The gallstone may become impacted in various locations throughout the GI tract. A review by Reisner and Cohen analyzing 1001 cases of gallstone ileus revealed that only 3.5% of stones were lodged in the duodenum, while 16.1% were found in the jejunum. The most common location, however, was the ileum, accounting for 60.5% of cases.[8]

This case highlights a rare presentation of gallstone ileus. While initial imaging and endoscopic findings suggested Bouveret syndrome due to a gallstone impacted in the duodenum (causing stomach dilation), surgical exploration revealed the stone had migrated distally to the jejunum. This indicates the patient was experiencing a "tumbling" or "rolling" obstruction phenomenon, where the obstructing stone spontaneously migrates distally, causing recurring bowel obstruction.[8]

Previously described methods to assist stone migration from the duodenum to avoid technically challenging surgery include milking the stone proximally for removal via gastrotomy, distally into the jejunum, or using high-pressure water flushes to propel the stone into the jejunum.[9,10] Furthermore, endoscopic lithotripsy to break up the gallstone in the duodenum and allow it to pass distally for later surgical retrieval has also been described.[11] However, none of these interventions were necessary in this particular instance. Purposefully waiting for spontaneous stone passage from the duodenum to the distal GI tract before surgical intervention has not been described in the current literature.

Another noteworthy aspect is the patient's familial history. Her daughter reportedly had gallstone ileus in 2020. While no documented cases of familial gallstone ileus exist in the current literature, this suggests a potential genetic predisposition. Typically, gallstone ileus is observed with a higher frequency in the elderly population[12] (60-84 years old[13]). Additionally, existing data showcases a female predominance in gallstone disease and gallstone ileus, ranging from 72% to 90%.[14,15] Consistent with this, a review of 128 cases by Cappell and Davis on Bouveret syndrome found the mean patient age of 74.1 ± 11.1 (SD) years (1–112) and a female-to-male sex ratio of 1.86.[16] Further investigation will help illuminate this rare cause of small bowel obstruction.

Conclusion

This case presents an unusual instance of gallstone ileus characterized by Bouveret syndrome and subsequent spontaneous distal migration of the obstructing gallstone, a phenomenon known as "tumbling" or "rolling" obstruction. It also highlights the previously unreported familial occurrence of gallstone ileus, suggesting a potential role for genetic predisposition in this condition. This case underscores the variable presentation of gallstone ileus and the importance of considering this diagnosis in patients with small bowel obstruction, even in the absence of typical risk factors.

Lessons Learned

This case underscores several important lessons for clinicians. First, gallstone ileus can exhibit a dynamic obstructive pattern, as the obstructing gallstone may undergo spontaneous migration within the gastrointestinal tract. Second, while Bouveret syndrome is characterized by duodenal impaction, the obstructing stone can subsequently migrate distally, necessitating careful evaluation of the entire GI tract. Third, a familial history of gallstone ileus may suggest a genetic predisposition to this condition, warranting further investigation into potential heritable factors. Finally, although gallstone ileus is more common

in elderly women, it should be considered in the differential diagnosis of small bowel obstruction in other patient populations.

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