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

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Introduction

- Gallstone ileus is a rare complication of gallstone disease and a rare cause of intestinal obstruction, accounting for only 1-4% of all bowel obstructions.[1]
- Gallstone ileus is caused by the impaction of a gallstone in the small bowel after passing through a biliary-enteric fistula, usually between the gallbladder and duodenum.[2]
- Gallstone ileus typically occurs after multiple bouts of cholecystitis as recurrent inflammation leads to fistula formation
- The treatment of choice is to remove the impacted stones either through an open or laparoscopic enterolithotomy. [3,4]
- While abdominal x-ray and ultrasound are typically the initial imaging modalities performed, they are insensitive to diagnosing gallstone ileus.
- Computed tomography (CT) is the imaging modality of choice for this disease.[5]
- Gallstone ileus in which a gallstone causes gastric outlet obstruction by lodging in the stomach or duodenum, is an even more rare complication of gallstone disease, known as Bouveret syndrome, and makes up only 1-3% of cases of gallstone ileus.[6,7]
- In Reisner and Cohen's review of 1001 cases of gallstone ileus, only 3.5% were found in the duodenum, 16.1% in the jejunum, and the majority, 60.5%, in the ileum.[8]

Case Report

A 74-year-old Caucasian female presented to the emergency department with a one-and-a-half-day history of nausea, vomiting, and severe epigastric abdominal pain. She had no significant medical history, but over the last two weeks, she had gradually worsening, burning epigastric pain. Her primary care physician trialed her on omeprazole, which did not help. Liver function tests were mildly abnormal (total bilirubin of 1.5 mg/dL, alkaline phosphatase 376 U/l, alanine aminotransferase 88 U/l, aspartate aminotransferase 38 U/l). White blood cell count was 16.8 x 10^9/l. CT scan (Figure 1 and 2) demonstrated a 2-cm rounded radiopaque structure within the second portion of the duodenum, obstructing the outflow of the stomach. The stomach was moderately dilated. A normal gallbladder was not seen. Mild intrahepatic biliary ductal dilation was most significant within the left hepatic lobe. The patient did not have any history of gallbladder resection.

This raised concern for possible Bouveret syndrome. The patient was admitted, and initial management included fluid resuscitation, nil per os (NPO), insertion of nasogastric tube, and appropriate analgesia and anti-nausea medications. The gastroenterology team attempted Esophagogastroduodenoscopy (EGD), which visualized an impacted gallstone in the second part of the duodenum (Figure 3). However, they could not visualize a cholecystoduodenal fistula due to the size and immobility of the stone. They were also unable to dislodge the stone or place a Roth net around it.

General surgery was then consulted for gastric outlet obstruction with concern for Bouveret syndrome. The patient consented to exploratory laparotomy with possible bowel resection and possible open cholecystectomy. Surgical intervention proceeded within 24 hours. There was no stone palpable within the proximal small bowel, but with further investigation, there was one palpable stone found within the jejunum (Figure 4). Enterotomy was made longitudinally, approximately 2 cm long, with electrocautery. One stone was delivered from the bowel lumen (Figure 5). The enterotomy was closed in an interrupted fashion with 3-0 silk. A falciform flap was created and placed 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 PACU in excellent condition. Her post-operative recovery was uneventful, and she was discharged home four days after her procedure.

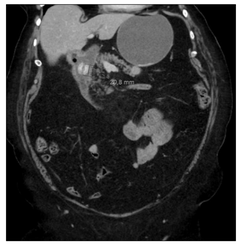


Figure 1: Coronal CT image demonstrates gallstone in a loop of small bowel in the right upper quadrant



Figure 2: Axial CT image demonstrates gallstone in a loop of small bowel in the right upper quadrant.

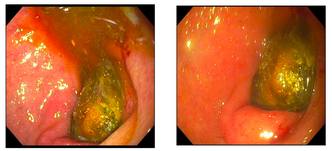


Figure 3: Gallstone impacted within the 2nd portion of the duodenum, identified via EGD

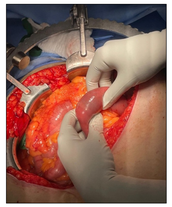


Figure 4: Gallstone identified in the proximal jejunum



Figure 5: Gallstone delivered via enterolithotomy

Discussion

- This case highlights a rare presentation of gallstone ileus:
  - The patient presented with Bouveret syndrome, as evidenced by her stomach dilation due to initial gallstone impaction in the duodenum on imaging and endoscopy.
  - However, on exploratory laparotomy, the stone was found within the jejunum.
  - This indicates that the patient was actively undergoing a tumbling phenomenon, whereby the obstructing stone continues to spontaneously migrate distally in the bowel, causing recurring bowel obstruction[8].
- While not necessary in this case, methods of physically assisting stone migration from the duodenum to avoid technically challenging surgery have previously been described:
  - Milking the stone proximally for removal via pyloroplasty or gastrostomy or distally into the jejunum or using high-pressure water flushes to push the stone into the jejunum[9,10]
  - Using endoscopic lithotripsy to break up a gallstone in the duodenum and allow it to pass distally for later surgical retrieval[11]
- This case is also notable for the familial history of gallstone ileus:
  - The patient's daughter reported having gallstone ileus as well
  - Upon review of current literature, there have been no case reports of familial occurrences of gallstone ileus
  - This indicates that inheritable genetic factors could predispose patients to gallstone ileus.
  - Typically, gallstone ileus is observed with a higher frequency in the elderly population,[12] usually between 60 - 84 years old.[13]
  - Along with the predominance of gallstone disease within female patients, gallstone ileus correlates with the female gender ranging from 72% - 90%.[14,15]
  - Similarly, in Cappell and Davis' review of 128 cases of Bouveret syndrome, the mean patient age reported in reviewed cases was 74.1 ± 11.1 (SD) years (1 - 112), and the female-to-male sex ratio was 1.86.[16]

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