

Bouveret's Syndrome with Severe Esophagitis and a Purulent Fistula

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Abstract

Bouveret's syndrome is a rare variant of gallstone ileus with an overall incidence of 1–3%. It is a rare cause of gastric outlet obstruction resulting from the passage and impaction of a large gallstone through a cholecystoduodenal fistula. A combination of diagnostic modalities is often required for a diagnosis. Management options include endoscopy and surgery. The most commonly performed procedures are enterolithotomy or gastrostomy, either alone or with cholecystectomy and fistula repair. We describe a unique variant of chronic Bouveret's syndrome with the unusual associations of severe esophagitis and a purulent fistula.

Introduction

Bouveret's syndrome is a cause of gastric outlet obstruction that results from the passage and impaction of a large gallstone through a cholecystoduodenal fistula. Bouveret first published 2 cases of this syndrome in 1896.¹ It tends to occur more commonly in women, has a mean age of 74 years, and has an overall incidence of 1–3%.² Various endoscopic and surgical techniques have been utilized in the management of Bouveret's syndrome, and a number of variants of the syndrome have been reported. We describe a unique variant: chronic Bouveret's syndrome characterized by the unusual associations of severe esophagitis and a purulent fistula.

Case Report

An 81-year-old Caucasian female with a history of gastroesophageal reflux disease (GERD) and atrial fibrillation was admitted to hospital with a 1-week history of worsening emesis and 2 days of inability to tolerate oral intake. She denied any sick contacts, fever, chills, diarrhea, melena, or abdominal pain. The patient related a similar episode 4 months prior that resolved spontaneously. Work-up at that time included a negative abdominal x-ray. Since then, she had reduced her meal sizes to avoid symptoms and had lost 30 lbs.

Her abdominal exam was unremarkable. Laboratory examination showed normal hemoglobin, white blood cell 20,000/uL, normal liver enzymes, and hypokalemic hypochloremic metabolic alkalosis. An abdominal x-ray showed pneumobilia with no small bowel obstruction. An abdominal computed tomography (CT) confirmed the pneumobilia and revealed a 2.5-cm outpouching in the duodenal bulb suspicious for duodenal diverticula or duodenal ulcer (Figure 1). Esophagogastroduodenoscopy EGD showed severe esophagitis (Figure 2) and a large gallstone completely obstructing the proximal duodenum with surrounding purulent material (Figure 3).

The area of obstruction was impassable endoscopically. Endoscopic lithotripsy was performed by breaking the stone and removing debris via forceps, water pick, snares, balloon sweeps, and a variety of nets. The scope was then advanced, and multiple pieces of stones fell into the distal duodenum. We noted a fistula extending

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Figure 1. Abdominal CT showing a 2.5-cm outpouching in the duodenal bulb suspicious for duodenal diverticula or duodenal ulcer.

into the gallbladder, and multiple stones were seen in the gallbladder. Endoscopic manipulation in the area caused several stones and purulent material to pass through the fistula into the duodenum. The quantity of stones led to aborting the procedure, starting piperacillin/tazobactam, and consulting surgery. At surgery, a gastrostomy allowed for the manual and complete removal of multiple remaining large gallstones. The fistula and gallbladder were left intact due to adhesions in the right upper quadrant and because the cystic duct was patent with no residual gallstones remaining. The patient made an uneventful recovery.

Discussion

Gallstone disease is a common digestive disease with a prevalence of 6% in men and 9% in women in the United States.³ Gallstone ileus is an unusual complication of cholelithiasis, occurring in less than 0.5% of patients with gallstones. Bouveret's syndrome is a rare variant and is responsible for 1–4% of all cases of mechanical obstruction.⁴ It is caused by the passage and impaction of a large gallstone through a cholecystoduodenal fistula that impacts in the proximal duodenum. After passing through the fistula, most of the gallstones pass asymptotically through the bowel. Impaction is most likely to occur in the ileum⁵ and less likely to occur in the duodenum and the proximal stomach.

A combination of diagnostic modalities is often required for diagnosis. An abdominal plain film in 30–35% of cases shows the Rigler triad (bowel obstruction, pneumobilia, and a calcified ectopic gallstone). Abdominal ultrasound and CT are the preferred noninvasive diagnostic tests for delineating the gastroduodenal anatomy, and can demonstrate a cholecystoduodenal fistula with ectopic stone location.² CT scans show the exact location of the ectopic stone and the site of obstruction and often visualize the biliary–enteric fistula.⁶ However, in 15–25% of patients, the stone is isoattenuating,

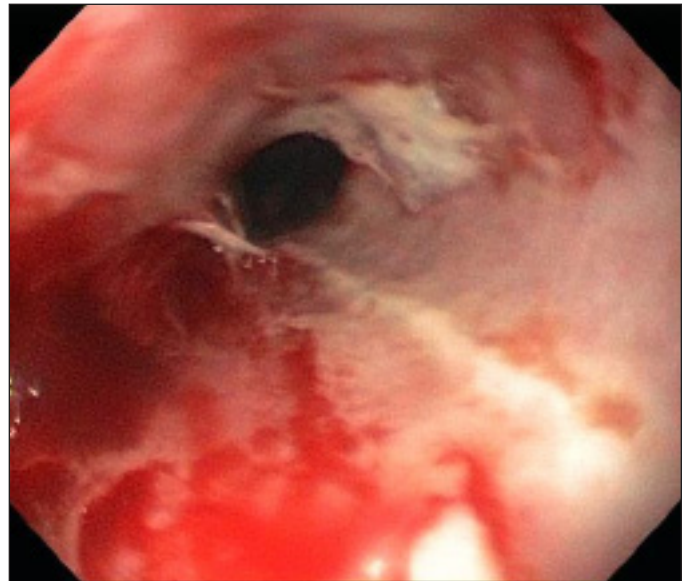


Figure 2. Severe esophagitis shown on EGD.

so differentiation of the stone from the surrounding bile and fluid is not possible. In such cases, magnetic resonance cholangiopancreatography (MRCP) can be helpful to distinguish isoattenuating stones from fluid and to visualize the fistula.⁷

The management of Bouveret's syndrome includes endoscopy and surgery. Endoscopic removal should be tried first because it is minimally invasive and has a low rate of complications.⁸ Many endoscopic techniques have been described, including mechanical lithotripsy, net extraction, electrohydraulic lithotripsy, intracorporeal laser lithotripsy, or combinations of these techniques.⁹ These techniques are



Figure 3. Large gallstone completely obstructing the proximal duodenum with surrounding purulent material.

more successful for proximal gallstone obstruction. The success rate of these time-consuming procedures is only 9%.¹⁰ Failed endoscopic attempts and distal migration of gallstones after dislodgement are indications for surgery. The most commonly performed procedures are enterolithotomy or gastrostomy, either alone or with cholecystectomy and fistula repair. A fistula may spontaneously close, and repair may be unnecessary if the cystic duct is patent and no residual gallstones are present.⁴

We suspect that this patient developed a partial obstruction approximately 4 months prior to our intervention. The prolonged partial obstruction led to significant weight loss and the development of esophagitis. The chronic course of this patient's illness is a unique presentation of Bouveret's syndrome. Perhaps the development of inflammation from the purulent fistula and elevated WBC count led to edema and complete obstruction. In published case reports of gallstone ileus and Bouveret's syndrome, patients present with acute symptoms of small bowel obstruction typically within 1 week. Weight loss in Bouveret's syndrome is reported in less than 20% of patients.² A PubMed review of 20 published cases and a comprehensive review of 128 cases by Cappel and Davis² revealed only 2% of Bouveret's syndrome cases presented with esophagitis and none of them had a purulent fistula.

Gallstone-related obstruction of the small bowel is a rare occurrence. Chronic Bouveret's syndrome is possible when an elderly patient presents with upper gastrointestinal obstruction symptoms, pneumobilia, significant weight loss, and esophagitis. In this patient, a purulent fistula and elevated WBC count was associated with the worsening of clinical symptoms. Antibiotics were instituted to prevent further complications associated with endoscopic instrumentation. Chronic Bouveret's syndrome should be added to the growing list of variants that have been reported in the literature.

Disclosures

Author contributions: All authors contributed equally to the manuscript. R. Bonam is the article guarantor.

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