

CASE REPORT

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Bouveret's syndrome as a rare cause of gallstone ileus: A surgical challenge

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ABSTRACT

Introduction: Bouveret syndrome, a rare cause of gastrointestinal outlet obstruction, was first described by Beassier in 1770. Afterward, this condition was named after Leon Bouveret in 1896. Bouveret syndrome is characterized by the passage of a gallstone through a cholecystogastric or more commonly through a cholecystoduodenal fistula, causing obstruction.

Case Report: A 70-year-old male patient without chronic degenerative diseases and with history of epigastric pain months prior to his current evaluation. Five days before hospital admission, the patient experienced worsening of the epigastric pain with an intensity of 8/10 accompanied by anorexia, vomiting, jaundice, and choluria. Physical examination with evidence of generalized jaundice ++, painful and distended abdomen in the upper right

quadrant. Diagnostic protocol was initiated based on preoperative tests, which showed hyperbilirubinemia of 2.78 mg/dL with an obstructive pattern and alteration of liver function tests. The hepatobiliary ultrasound showed a lithiasic sclero-atrophic gallbladder, a common bile duct diameter of 5 mm and positive sonographic murphy sign. The patient was taken to the operating room where, after general anesthesia and in a supine position, a 13 mmHg pneumoperitoneum was created and laparoscopic working ports were introduced. A laparoscopic cholecystectomy + transcystic cholangiography + resection of the cholecystoduodenal fistula were performed.

Conclusion: A high degree of clinical suspicion and imaging studies are required to establish Bouveret syndrome diagnosis, nevertheless in 50% of cases, the diagnosis is made intraoperatively. Early diagnosis and adequate surgical intervention are essential for a favorable prognosis.

Keywords: Cholecystoduodenal fistula, Cholelithiasis, Gallstone ileus, Pneumobilia

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INTRODUCTION

Bouveret syndrome, a rare cause of gastrointestinal outlet obstruction, was first described by Beassier in

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1770. Afterward, this condition was named after Leon Bouveret in 1896, who presented two cases of patients with this condition. Bouveret syndrome is caused by the impaction of a gallstone into the pylorus or duodenum and is characterized by the passage of a gallstone through a cholecystogastric or more commonly, through a cholecystoduodenal fistula, causing obstruction of the gastric outlet. Cholelithiasis is a common disease which can lead to several complications. Bouveret syndrome accounts for 0.3% of complications from cholelithiasis, and gallstone ileus accounts for approximately 1–3% of cases of mechanical gastrointestinal obstruction. The typically affected population is the elderly, with a median age of 70 years old, which contributes to the high mortality rates estimated to be 12–30% [1, 2, 3, 4].

CASE REPORT

A 70-year-old male patient without chronic degenerative diseases and with history of epigastric pain months prior to his current evaluation. Five days before hospital admission, the patient experienced worsening of the epigastric pain with an intensity of 8/10 accompanied by anorexia, nausea, vomiting, as well as jaundice and choloria. Physical examination with evidence of generalized jaundice ++, painful and distended abdomen in the upper right quadrant with no evidence of peritoneal irritation.

Diagnostic protocol was initiated based on preoperative tests, which showed leukocytes of 7.33, platelets of 254,000, total bilirubin of 2.78 mg/dL, direct bilirubin of 1.56 mg/dL, indirect bilirubin of 1.22 mg/dL, aspartate aminotransferase of 126 U/L, alanine aminotransferase of 333 U/L, gamma glutamyl transpeptidase of 749 U/L, alkaline phosphatase of 144 U/L, and lactate dehydrogenase of 233 U/L. The hepatobiliary ultrasound showed a lithiasic sclero-atrophic gallbladder, a common bile duct diameter of 5 mm and positive sonographic murphy sign.

The patient was taken to the operating room where, after general anesthesia and in a supine position, a 13 mmHg pneumoperitoneum was created and laparoscopic working ports were introduced. During diagnostic laparoscopy a cholecystoduodenal inflammatory process was identified, as well as, a cholecystoduodenal fistula (Figure 1). The fistula was dissected, followed by the dissection of the Calot's triangle, dissecting the cystic duct and performing transcystic bile duct exploration, identifying a stone within. A balloon catheter was introduced and gallstones were extracted (Figure 2). A transcystic cholangiography was performed, showing adequate passage into the duodenum and into the right, left, and common hepatic ducts (Figure 3). The balloon catheter was removed, the cystic duct and artery were stapled and the gallbladder was dissected. Then, the cholecystoduodenal fistula was stapled with a tri-staple stapler and reinforced with mechanical

sutures (Figure 4). Finally, hemostasis was confirmed, a thorough cleaning was performed, a Penrose drain was placed in Winslow's hiatus and the gallbladder was extracted. A laparoscopic cholecystectomy + transcystic cholangiography + resection of the cholecystoduodenal fistula were performed. The patient presented favorable clinical evolution and was discharged three days after surgery.

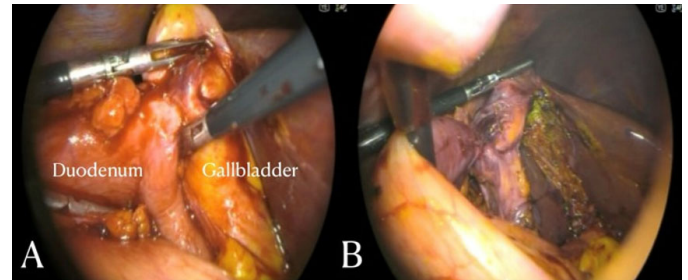


Figure 1: Inflammatory process and cholecystoduodenal fistula.

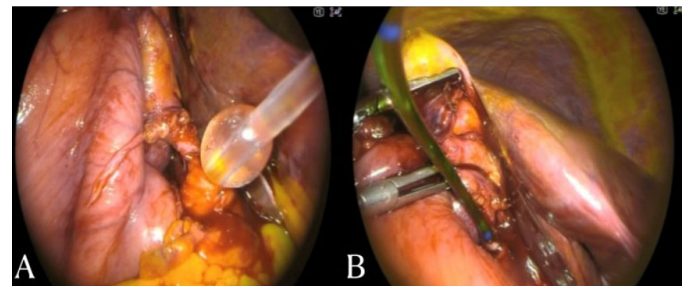


Figure 2: (A) Balloon catheter for gallstone extraction, (B) placement of cholangiography probe.

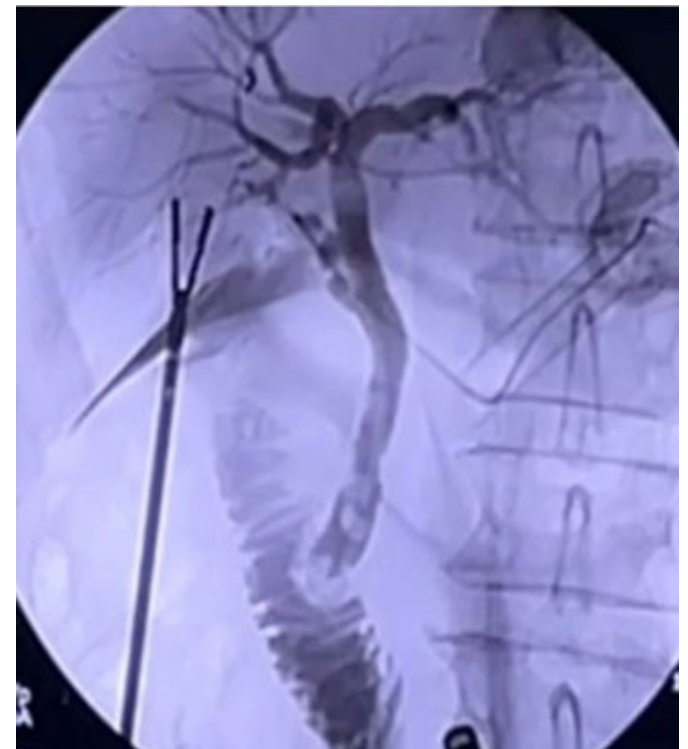


Figure 3: Transcystic cholangiography with adequate passage into the duodenum and into the right, left, and common hepatic ducts.

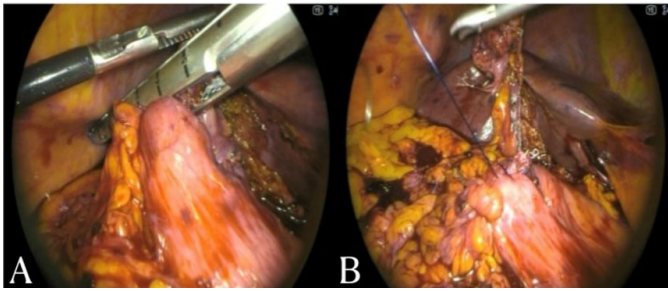


Figure 4: (A) Stapling of the cholecystoduodenal fistula with a tri-stapler, (B) reinforcement of the staple line with mechanical suture.

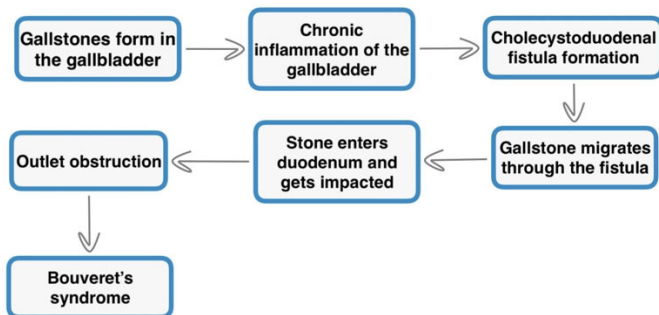


Figure 5: Pathophysiology of bilioenteric fistula formation and Bouveret syndrome.

DISCUSSION

The inflammation and adherence of the gallbladder to the pylorus or duodenum are a consequence of multiple episodes of acute cholecystitis which can lead to an ischemic tear of the gallbladder and enteric wall, in addition to the mechanical tension exerted by the gallstone, all favoring the formation of a bilioenteric fistula (Figure 5). Alternatively, there have been reports of the development of bilioenteric fistulas secondary to gallbladder malignancy. Bilioenteric fistulas most commonly present as cholecystoduodenal fistulas, representing 68% of all bilioenteric fistulas, followed by cholecystocolic fistulas with 17% and lastly, by cholecystogastric fistulas, representing 5% of all cases, which is the rarest due to gastric wall thickness [1, 4, 5].

Bouveret's syndrome risk factors include being elderly with a median age of 70 years, female sex, gallstones between 2 and 8 cm, and gastrointestinal anatomical alterations. The clinical presentation of Bouveret syndrome is nonspecific, which delays the diagnosis and contributes to high morbidity and mortality, in addition to associated comorbidities. In most cases in which a bilioenteric fistula is present, gallstones are eliminated through vomiting or defecation, and therefore, symptoms arise with larger stones. The most common symptoms are nausea, vomiting, hematemesis, weight loss, abdominal pain and anorexia. Hematemesis results from a Mallory–Weiss tear and in some cases emesis can be severe enough that may lead to a Boerhaave syndrome. Physical examination may reveal abdominal distension and tenderness, as well as altered bowel

movements. Bouveret's syndrome complications include biliary or duodenal perforations, biliary infection, acute pancreatitis, cancer, and recurrent Bouveret syndrome [1, 4, 5, 6, 7].

Regarding the diagnosis, laboratory tests are non-specific. Bouveret syndrome is mainly diagnosed by imaging studies. Rigler's triad, which consists of pneumobilia, an ectopic gallstone, and distended stomach, is pathognomonic, although it only appears in 35% of the cases. Abdominal ultrasound can also demonstrate Rigler's triad. Esophagogastroduodenoscopy provides direct visualization of the gallstones and has the additional advantage of being therapeutic, however, in almost 33% of the cases, it fails to visualize the gallstone. The best diagnostic method is the contrast-enhanced computed tomography, which has a sensitivity of 90% and specificity of 100% in detecting gallstone ileus. Abdominal computed tomography (CT) can provide more detailed information about the fistulous tract, and the ectopic calculus, and may help identify complications [4, 7, 8].

Treatment options include endoscopic, laparoscopic, or open disimpaction of the stone. The endoscopic approach, diagnostic and potentially therapeutic, is preferred due to the lowest mortality; nonetheless, its effectiveness depends on the size of the gallstone and on adequate endoscopic skills. Endoscopic modalities include nets, baskets, and lithotripsy techniques such as mechanical, electrohydraulic, and intracorporeal lasers which can be used prior to endoscopic extraction. Complications of endoscopic treatment include impaction of the stone in the esophagus, gastrointestinal bleeding, and perforation [2, 6, 9].

Although the endoscopic approach is preferred, surgery will be required in 90% of cases. There are currently 3 surgical approaches. Laparoscopic or open enterotomy with gallstone extraction and spontaneous closure of the fistula, which should be offered in elderly patients with associated comorbidities. A one stage procedure involving enterotomy and gallstone extraction, with or without cholecystectomy and repair of the fistula, which should be considered in patients with good general condition. The last one is the two stage procedure consisting of enterotomy with gallstone extraction followed by an elective cholecystectomy and fistula repair after 4–6 weeks. Cholecystectomy is indicated to prevent recurrence and biliary complications, and only 10% of the patients will require delayed cholecystectomy with fistula repair for persistent symptoms. Laparoscopic surgery is a safe and an effective alternative to open surgery for gallstone ileus; however, it is used in only 10% of surgical managed gallstone ileus cases with a high conversion rate to laparotomy [2, 3, 6].

CONCLUSION

Due to the non-specific symptoms and low incidence of the syndrome, a high degree of clinical suspicion

and imaging studies are required to establish Bouveret syndrome diagnosis, nevertheless, in 50% of cases the diagnosis is made intraoperatively. Early diagnosis and adequate surgical intervention are essential for a favorable prognosis. The endoscopic approach remains the first-line treatment, particularly in the elderly with comorbidities. Alternatively, the laparoscopic approach and repair of the fistula remains a complex and controversial issue among surgeons.

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Carlos Tadeo Perzabal Avilez – Conception of the work, Analysis of data, Interpretation of data, Revising the

work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Saul Jasam Ruiz Cereceres – Conception of the work, Analysis of data, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

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Conflict of Interest

Authors declare no conflict of interest.

Data Availability

All relevant data are within the paper and its Supporting Information files.

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