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Case Report

Bouveret syndrome secondary to a cholecystoduodenal fistula in a patient with locally advanced cholangiocarcinoma

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ABSTRACT

Bouveret syndrome is a rare form of gallstone ileus characterized by gastric outlet obstruction caused by the impaction of a large gallstone that migrates through a biliary-enteric fistula. Its occurrence in patients with active biliary malignancy is exceptionally uncommon and poses significant diagnostic and therapeutic challenges. We report the case of a 63-year-old woman with locally advanced, unresectable cholangiocarcinoma who presented with acute abdominal pain, hemodynamic instability, and severe metabolic derangement during first-line systemic therapy. Computed tomography revealed a contained gallbladder perforation with a cholecystoduodenal fistula and a large gallstone impacted in the duodenal bulb, causing secondary gastric dilatation consistent with Bouveret syndrome. Due to clinical deterioration and unfavorable local inflammatory conditions, surgical management was selected. Exploratory laparotomy was performed, and the stone was mobilized retrogradely into the stomach, followed by anterior gastrotomy and successful extraction. The management of Bouveret syndrome remains controversial, particularly in patients with advanced malignancy, as endoscopic treatment has limited success in cases involving large stones or severe inflammation. In this patient, stone size, local anatomy, and oncologic status justified a palliative surgical approach focused on symptom relief and damage control. Bouveret syndrome in the setting of advanced cholangiocarcinoma is rare and associated with poor prognosis; however, individualized, multidisciplinary decision-making is essential, and surgical intervention remains a valid option in unstable patients or when endoscopic management is not feasible.

Keywords: Bouveret syndrome, Gallstone ileus, Cholangiocarcinoma, Cholecystoduodenal fistula, Surgery

INTRODUCTION

Bouveret syndrome is a rare form of gallstone ileus characterized by mechanical obstruction of the gastric outlet, most commonly at the duodenal level, caused by the impaction of a large gallstone that migrates through a cholecystoduodenal fistula.

It accounts for approximately 1-3% of cases of gallstone ileus and is typically observed in elderly patients with long-standing gallstone disease.[1-4]

The clinical presentation is often nonspecific and may include nausea, vomiting, epigastric pain, and signs of gastric outlet obstruction, frequently leading to delayed diagnosis. Computed tomography is considered the imaging modality of choice, as it allows identification of the ectopic gallstone, pneumobilia, and the biliary-enteric fistula.[2,3]

Management strategies vary depending on patient comorbidities and anatomical findings, ranging from endoscopic stone extraction to surgical intervention, with

surgery remaining the definitive treatment in complicated or refractory cases.[3,4]

Its occurrence in the setting of locally advanced cholangiocarcinoma is exceptionally rare, with only isolated cases reported in the literature, posing significant diagnostic and therapeutic challenges.[5,6]

We report the case of a 63-year-old woman with unresectable cholangiocarcinoma who developed acute Bouveret syndrome secondary to a cholecystoduodenal fistula, requiring urgent surgical intervention.

CASE REPORT

A 63-year-old woman (BMI 24.15) with no family history of malignancy presented with a medical history significant for type 2 diabetes mellitus, mixed dyslipidemia, chronic venous insufficiency, and a prior episode of femoral vein thrombosis secondary to COVID-19 infection in 2020. She had no relevant surgical history.

In June 2024, she developed obstructive jaundice, choluria, and an unintentional weight loss of 20 kg. Initial diagnostic workup, including abdominal ultrasound and magnetic resonance imaging, was performed at a secondary-level care hospital. These studies reportedly demonstrated intra- and extrahepatic biliary dilation, proximal common bile duct stenosis secondary to a 16-mm lesion in the pancreatic head, and a 36-mm gallstone within the gallbladder. However, the original imaging studies could not be retrieved for inclusion in this case report, as they were performed at an outside institution and were not available for review during the manuscript preparation process.

Upon her arrival at our tertiary care center, further diagnostic and therapeutic evaluation was performed. Endoscopic retrograde cholangiopancreatography was performed with biliary stent placement (Figure 1).

Endoscopic ultrasound-guided biopsy confirmed poorly differentiated biliary adenocarcinoma (HER2 1+, pMMR), with an initial CA 19-9 level of 1,884 U/mL (reference range: <37 U/mL). Histopathological images were not available for inclusion in this case report. Following multidisciplinary evaluation by the hepatopancreatobiliary surgery team, the tumor was deemed unresectable due to arterial involvement related to an anatomical vascular variant.

The patient initiated first-line systemic therapy. After seven cycles, she presented with acute abdominal pain, hemodynamic instability, and severe glycemic decompensation. Computed tomography revealed a contained gallbladder perforation with formation of a cholecystoduodenal fistula and impaction of a 3×5 cm gallstone in the duodenal bulb, causing secondary gastric dilation consistent with Bouveret syndrome (Figure 2).

An exploratory laparotomy was performed. Due to severe local inflammation, distal enterotomy was not feasible. The gallstone was mobilized retrogradely into the stomach, and an anterior longitudinal gastrotomy was performed for extraction, followed by primary closure reinforced with an omental patch. The gross appearance of the extracted gallstone confirmed its large size, measuring approximately 3×5 cm (Figure 3).

Postoperative recovery was complicated by nosocomial pneumonia and recurrent episodes of acute cholangitis due to Enterococcus spp. In October 2025, the patient developed pericholangitic abscesses and refractory septic shock. Given the poor prognosis, comfort-focused care was initiated following family consensus. The patient died in November 2025.

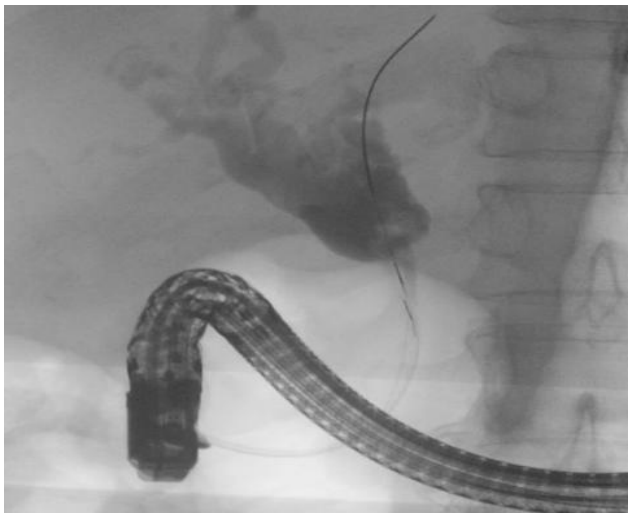

Figure 1: Endoscopic retrograde cholangiopancreatography with biliary stent placement.

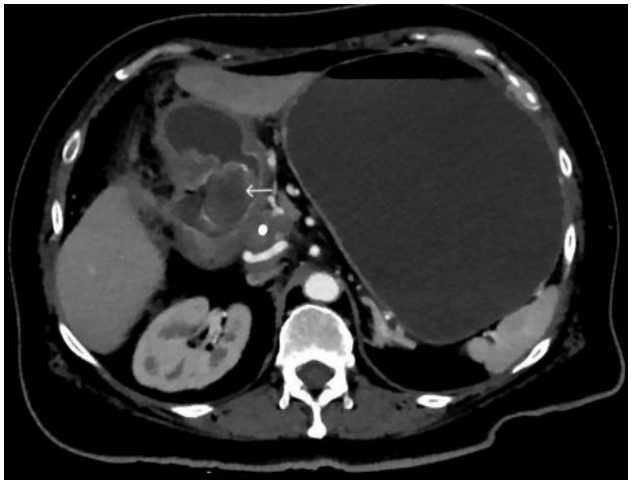

Figure 2: Contrast-enhanced axial abdominal computed tomography demonstrating a large gallstone impacted in the duodenal bulb (arrow), associated with marked gastric dilation, consistent with Bouveret syndrome.

Figure 3: Gross appearance of the extracted gallstone measuring approximately 3×5 cm after surgical removal via anterior gastrotomy.

DISCUSSION

Bouveret syndrome is an exceptionally rare presentation of gallstone ileus, accounting for approximately 1-3% of cases, and is defined by gastric outlet obstruction caused by migration of a gallstone through a bilioenteric fistula, most commonly cholecystoduodenal, with subsequent impaction in the pylorus or duodenum.[1-3] It predominantly affects elderly patients with multiple comorbidities and is typically associated with chronic gallbladder inflammation and recurrent cholecystitis, which promote adhesion and erosion between the gallbladder and adjacent bowel.[2-4] Although Bouveret syndrome has been reported in association with biliary malignancies, this scenario remains uncommon and has been described mainly in isolated case reports, often in patients with advanced disease.[5,6]

Clinically, patients usually present with nonspecific symptoms such as nausea, vomiting, epigastric pain, and intolerance to oral intake, which may delay diagnosis, particularly in individuals with complex oncologic backgrounds. In the present case, these manifestations were initially obscured by symptoms related to extrahepatic cholangiocarcinoma, complicating early recognition of gastric outlet obstruction.

In the present case, the clinical course was initially dominated by extrahepatic cholangiocarcinoma. Vascular involvement is a central determinant of resectability in cholangiocarcinoma, and contemporary surgical frameworks emphasize that extensive arterial or portal vein invasion may preclude curative resection when safe reconstruction or adequate future liver remnant cannot be ensured.[7-9] Accordingly, detailed preoperative vascular assessment is essential not only for technical planning but also for defining overall oncologic strategy in hepatopancreatobiliary malignancies.[8,9]

The occurrence of Bouveret syndrome during first-line systemic therapy represents a major complication in an already high-risk clinical setting. While bilioenteric fistulas are classically associated with benign gallstone disease, their formation has been described as a consequence of sustained inflammation, pressure-related ischemia, and erosion of the gallbladder wall into adjacent bowel.[3,10] From a diagnostic standpoint, computed tomography plays a pivotal role, as it allows simultaneous identification of gastric outlet obstruction, ectopic gallstone, pneumobilia, and the presence of a bilioenteric fistula, even in patients with distorted anatomy due to malignancy or prior interventions. In parallel, outcomes in advanced biliary tract cancer have shown modest improvement with contemporary first-line regimens combining gemcitabine and cisplatin with immune checkpoint inhibition, allowing disease control in selected patients treated with palliative intent.[11,12]

Management of Bouveret syndrome remains heterogeneous. Endoscopic therapy is frequently considered as an initial approach in selected patients; however, systematic reviews indicate limited success rates, particularly in cases involving large stones or firm duodenal impaction.[13] Stone size greater than 2.5-3 cm has consistently been identified as a predictor of endoscopic failure and the need for operative intervention.[2,13] In unstable patients or when endoscopic treatment is unlikely to succeed, surgical management is generally required, with operative strategies tailored to minimize physiologic stress and avoid extensive dissection in severely inflamed fields.[4,14]

Intraoperatively, marked inflammation may preclude safe duodenotomy or enterotomy. Retrograde mobilization of the stone into the stomach followed by gastrotomy has been described as a feasible alternative in hostile anatomy, permitting resolution of obstruction while limiting dissection around the duodenum and the fistulous area.[15]

Postoperative morbidity in patients with Bouveret syndrome remains substantial, with reported mortality rates higher in elderly individuals with comorbidities, sepsis, or when complex one-stage procedures are performed.[4,14] In the present case, overall survival from symptom onset should be interpreted in the context of the known survival benefit associated with modern first-line systemic therapy for advanced biliary tract cancer, which has demonstrated clinically meaningful disease control in selected patients.[11,12] This case highlights the importance of multidisciplinary decision-making in advanced hepatopancreatobiliary disease and supports surgical management as an appropriate option when endoscopic therapy is unlikely to succeed or when urgent intervention is required due to clinical instability.[13,14]

CONCLUSION

Bouveret syndrome in the setting of active cholangiocarcinoma represents an exceptionally rare and poor-prognosis condition that requires individualized,

multidisciplinary decision-making. In this case, stone size, severe inflammation, and unfavorable vascular anatomy precluded curative intent and justified a palliative surgical approach focused on damage control and relief of mechanical obstruction. Despite a complicated postoperative course and disease progression, prolonged survival relative to tumor aggressiveness was achieved, suggesting a clinical benefit from combined systemic therapy. Early recognition of this rare entity and careful selection of therapeutic strategies remain essential in advanced hepatopancreatobiliary disease.

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