



Case Report

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A Rare Case of Bouveret Syndrome Following Subtotal Cholecystectomy



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Abstract

Background

Bouveret syndrome is an uncommon cause of gastric outlet obstruction wherein a large gallstone becomes lodged in the proximal duodenum or pylorus through a cholecystoduodenal fistula. Its incidence is extremely rare in patients who underwent prior subtotal cholecystectomy.

Case presentation

We report a case of a 54-year-old Caucasian lady presenting with progressive vomiting, intolerance of solids and liquids and abdominal distension 6 years after a subtotal cholecystectomy. Cross-sectional imaging revealed gastric dilatation, a gallstone in residual gallbladder tissue and proximal duodenal obstruction. As conservative as well as endoscopic attempts failed to resolve the resultant gastric outlet obstruction, she was referred to the local hepato-biliary center who managed her with a one stage completion cholecystectomy, full closure of the cholecystoduodenal fistula with a falciform patch and gastrojejunostomy. She had an uneventful recovery and remained symptom-free in 2 months follow-up.

Discussion

This case highlights the need to consider Bouveret Syndrome as differential diagnosis in patients who presents with gastric outlet obstruction after subtotal cholecystectomy. Delayed complications from remnant gallbladder tissue should remain within the differential and cross-sectional imaging is an important diagnostic tool. Surgical intervention, which addresses the obstructing stone as well as the fistula, can lead to more favorable outcomes when conservative and endoscopic measures fail.

Conclusion

Clinicians should consider Bouveret syndrome as a potential rare long-term complication after a subtotal cholecystectomy. Effective management of this unique patient population requires early diagnosis and a multidisciplinary approach.

Keywords: Bouveret Syndrome; Gastric Outlet Obstruction; Subtotal Cholecystectomy; Cholecystoduodenal Fistula

Abbreviations: OGD: Oesophago-Gastro-Duodenoscopy; NG: Nasogastric; NJ: Nasojejunal; US: Ultrasound

Background

Bouveret Syndrome is a rare cause of gastric outlet obstruction due to gallstone disease [1]. It occurs when a large stone becomes impacted in the proximal duodenum or pylorus due to the formation of fistula between the gallbladder and duodenum [2]. The condition was first reported in 1770 by the French surgeon Leon Bouveret who published two case reports in 1896 [3]. Gallstone ileus, which accounts for less than 4% of bowel obstruction, is generally rare. Bouveret Syndrome is even less common, accounting for only 1-3% of gallstone ileus cases [4]. To our best knowledge, this is the first case of Bouveret Syndrome reported in English language in patient with prior subtotal

cholecystectomy. A detailed review of the published English language literature by means of a comprehensive electronic search of MEDLINE and manual review of the bibliographies of relevant papers failed to identify any other previously documented similar presentation. In the case presented, the patient had a history of subtotal cholecystectomy performed 6 years before developing Bouveret Syndrome.

Case Presentation

A 54-year-old Caucasian lady presented to James Cook University Hospital Accident and Emergency (A&E) in 2024 in

United Kingdom (UK) with a three-week history of worsening burping, vomiting, and reduced bowel movements. She reported an inability to tolerate both solids and liquid. On physical examination, she exhibited mild tachycardia, epigastric distension but her abdomen was otherwise soft and non-tender. Laboratory investigations showed a white cell count of $9.7 \times 10^9/L$, total bilirubin of 30 mmol/L, alanine aminotransferase of 170 U/L and C - Reactive Protein (CRP) of 22 mg/L.

Her previous medical history includes gout, IBS, supra-umbilical hernia repair, and subtotal cholecystectomy in June 2018.

Cross-sectional imaging (Figure 1) showed significant proximal duodenal and gastric dilatation and a remnant gallbladder containing a large calculus with associated duodenal stricture. Oesophago-gastro-duodenoscopy (OGD) revealed obstruction at the D1/D2 level and inability to visualize the lumen (Figure 2), with solid food residue in the stomach. A Nasojejunal tube was placed following balloon dilatation to assist with enteral feeding. Biopsy from the duodenum during OGD showed no evidence of dysplasia or malignancy. Based on clinical presentation, imaging and endoscopic findings, a diagnosis of Bouveret Syndrome following subtotal cholecystectomy was made.

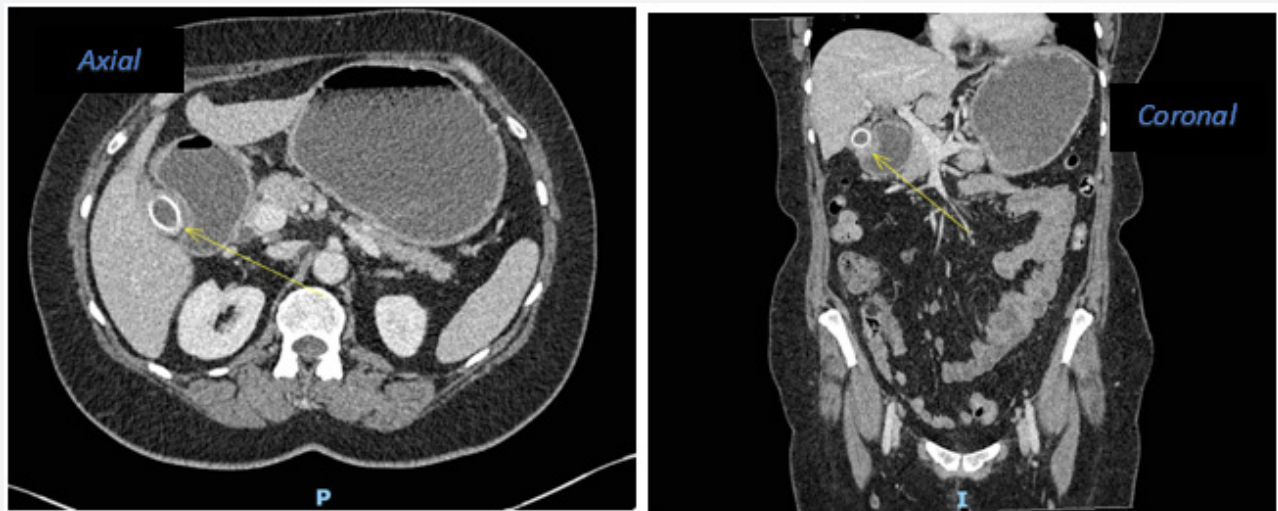


Figure 1: Axial and coronal views of the CT abdomen and pelvis with contrast. A yellow arrow shows a radiolucent stone within the gallbladder remnant which causes extrinsic compression of duodenum with associated duodenal and stomach dilatation.

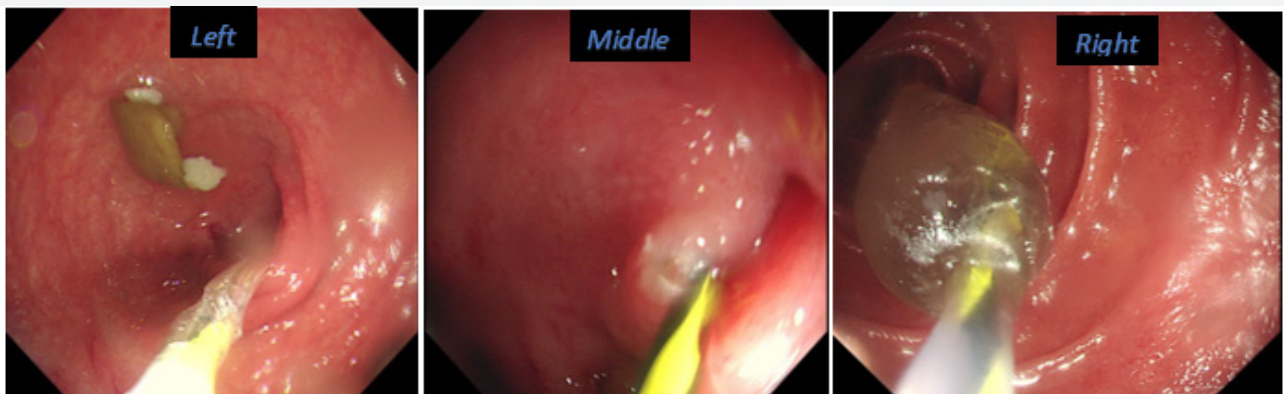


Figure 2: OGD from left to right showed patent pylorus (left picture), tight narrowing at the junction between the first (D1) and second (D2) portion of the duodenum which could not accommodate a 12Fr NJ tube (middle picture). Following balloon dilatation (right picture), it was possible to advance into D2.

Treatment

Initially, the patient was managed conservatively with nasogastric (NG) tube decompression of the stomach, intravenous

fluids and nutritional support via nasojejunal (NJ) tube. Due to recurrent symptoms, dislodgements of the NJ tube and subsequent inability to re-site the feeding tube even under fluoroscopic guidance, the patient was referred to the tertiary

hepato-biliary unit for definitive management. Subsequent management at tertiary center included endoscopic ultrasound assessment, further biopsies and repeat cross-sectional imaging in the form of CT scan. Once the diagnosis was re-confirmed and, more importantly other pathologies excluded, the patient underwent elective laparotomy, completion cholecystectomy, closure of cholecystoduodenal fistula with falciform patch and gastrojejunostomy in tertiary center 3 months after her original presentation.

Outcome and follow up

The patient had an uneventful post-operative course: oral diet was gradually built up, and enteral feeding weaned off and she was discharged home on day 6 post-operatively. 2 months follow-up showed the patient was symptoms free and managing to gain weight.

Discussion

Bouveret Syndrome presents a diagnostic and therapeutic challenge. This rare form of gallstone ileus, accounting for only 1-3% of all cases of gallstone ileus [5], occurs when a large gallstone erodes through the gallbladder wall, creating a cholecystoduodenal fistula, and subsequently obstructing the gastric outlet or proximal duodenum. The classic Rigler's triad, while helpful, is only present in a minority of cases (around 20-50%) and includes pneumobilia (air in the biliary tree), small bowel obstruction and an ectopic gallstone. Risk factors include advanced age, female gender, and a history of gallstone disease or gallstone size greater than 2.5cm. Early diagnosis and intervention are crucial to improve patient outcomes. Diagnosis is often delayed due to the non-specific nature of the symptoms. These include nausea, vomiting, abdominal pain and distension, which can mimic other gastrointestinal disorders. Furthermore, the intermittent nature of the obstruction can further delay diagnostic process [6].

CT scan is the gold standard for diagnosing gallstone ileus, with reported sensitivity of 93% and specificity of 100% for detecting ectopic gallstones and associated complications [7]. Ultrasound (US) is non-invasive and inexpensive, but its sensitivity ranges from 60-88% and specificity from 87-98% for detecting gallstones; however, its utility is limited if bowel gas obscures visualization or if the stone is located distally. Magnetic Resonance Cholangiopancreatography (MRCP) offers detailed biliary imaging, with sensitivity and specificity both generally above 90% for biliary tree abnormalities and fistulas [8], though it is less sensitive than CT for bowel obstruction itself. While CT is the primary modality due to its high accuracy, US and MRCP provide valuable complementary information depending on availability and the clinical scenario.

Endoscopic removal of gallstones causing obstruction in Bouveret Syndrome can involve several techniques beyond mechanical lithotripsy. These include electrohydraulic lithotripsy

(using shock waves), laser lithotripsy, extracorporeal shock wave lithotripsy (external shock waves), balloon extraction, retrieval baskets or snares and direct manual advancement of the stone. The choice of technique depends on the size, location and characteristics of the stone as well as the available equipment and expertise. If endoscopic methods fail, surgery may be required [9]. Surgical approach in patients who still have intact gallbladder typically involves enterolithotomy (stone removal) with or without cholecystectomy and fistula repair. The decision depends on patient factors and surgical expertise [10].

Since our patient had undergone subtotal cholecystectomy 6 years earlier, the surgical management in this case included a laparotomy with completion cholecystectomy, closure of the cholecystoduodenal fistula using a falciform ligament patch, and creation of a gastrojejunostomy. These steps were taken after conservative and endoscopic treatments failed to resolve the patient's obstruction. This approach addresses both the acute obstruction and the underlying cause, aiming to prevent recurrence and improve long-term outcomes. Treatment at tertiary center addressed all aspects of the disease in one stage operation: removal of the obstructing stone, closure of the fistula, and correction of gastric outlet obstruction. However, this approach is seen as more invasive, carries a higher immediate operative risk (especially in elderly or frail patients), and is technically challenging in inflamed or scarred fields. Therefore, in this case it was carried out in a high volume hepato-biliary center.

The alternative is a two-staged approach, which is often reserved for high-risk or frail patients. The initial enterolithotomy step relieves obstruction; and after the patient recovers and stabilizes, a second procedure (weeks to months later) may be done to repair the fistula and perform cholecystectomy if required. This method's main advantage is a quicker, less invasive and thus ideal for those not fit for prolonged operations. However, not all patients complete the second stage, leaving the fistula and gallbladder in place, which increases the risk of recurrence or persistent biliary symptoms [11]. Several previous studies have suggested that cholecystectomy and fistula repair in patients who still have the gallbladder should be delayed as performing these procedures immediately can lead to considerable risks of complications and death [11]. However, in this case, because the patient was relatively young and did not have other co-morbidity, an enterolithotomy along with completion cholecystectomy, fistula repair and gastric bypass was carried in one stage procedure without any complications at 2 months follow up.

Conclusion

This case highlights an exceptionally rare presentation of Bouveret Syndrome occurring 6 years after subtotal cholecystectomy. While Bouveret Syndrome itself is an uncommon cause of gastric outlet obstruction, its development in a patient with remnant gallbladder tissue is even rarer. As shown in this case, clinicians should consider Bouveret Syndrome as a potential

rare long-term complication after subtotal cholecystectomy and effective management of this unique subset of patients requires early diagnosis and a multidisciplinary approach.

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