

Case Report

Concomitant Bouveret's syndrome and gallstone ileus: case report and literature review

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ABSTRACT

Gallstone ileus and Bouveret's syndrome are rare gallstone complications; therefore, their combination is even rarer. The delay of diagnosis is associated with high rates of morbidity and mortality. We reported a 65-year-old woman presented with a three-day history of epigastric and lower right abdominal pain, nausea, and vomiting. A radiopaque shadow was noted on an abdominal X-ray in both the right upper and lower quadrants. In addition to the cholecystoduodenal fistula, the CT abdomen revealed a combined ileal gallstone ileus with small bowel dilatation and stone in the second portion of the duodenum (Bouveret's syndrome). Initial attempts to extract duodenal stones endoscopically were unsuccessful. Therefore, the patient underwent an exploratory laparotomy and removal of both ileal and duodenal stones. The patient had an uneventful postoperative course. Serious complications include obstruction of the gastric outlet or the small intestine due to gallstone ileus, Bouveret's syndrome, or both. Despite the high failure rate, endoscopic extraction of the stone alone or in combination with lithotripsy is the standard treatment for Bouveret's syndrome. Furthermore, surgical intervention is the standard treatment for gallstone ileus. Gallstone ileus and Bouveret's syndrome remain diagnostic and therapeutic challenges, which necessitate an immediate multidisciplinary approach to prevent substantial morbidity and mortality. Further reports or larger-scale studies are encouraged.

Keywords: Gallstone ileus, Bouveret syndrome, choleduodenal fistula

INTRODUCTION

Gallbladder stones are a common health problem; by age 75 years, approximately 35% of females and 20% of males have developed cholelithiasis.¹ However, the increase in people's interest in weight loss led to an increase in the use of rapid weight loss procedures, which resulted in an increase in the incidence of gallbladderstones among younger generations, with a prevalence of 24.7%.²

Cholecystoenteric fistula (CEF) is a rare complication of cholelithiasis, occurring in about 6% of patients.³ The CEF formation can be caused by pressure necrosis and compression of a gallbladder stone against the gallbladder wall, followed by passage of stones through the fistula

tract to the proximal duodenum or pylorus (1-3%), resulting in gastric outlet obstruction (GOO, Bouveret's syndrome) or to the terminal ileum (84%), resulting in small bowel obstruction (SBO, gallstone ileus).^{1,3}

Gallstone ileus (GSI) is a rare complication of gallbladder stones that affects 0.3% to 0.5% of cholelithiasis patients, most of whom are elderly females.³ GSI accounts for approximately 5% of mechanical SBO, whereas approximately 320 cases of Bouveret's syndrome have been reported worldwide.^{4,5} To the best of our knowledge, both conditions are rare, while the simultaneous occurrence of both is extremely rare worldwide. We are consequently the first to report this case from Saudi Arabia and the Arab Gulf States.

CASE REPORT

A 65-years-old female, a known case of type II diabetes mellitus and hypertension, presented to the emergency department with a three-day history of epigastric and right lower quadrant abdominal pain, nausea, and vomiting. There was no fever, additional gastrointestinal or urinary manifestations.

The patient has previously been hospitalized twice for acute calcular cholecystitis and has refused surgical intervention (laparoscopic cholecystectomy).

On examination, vital signs were within normal ranges. The abdomen was soft but mildly distended with hyperaudible bowel sounds. Blood tests revealed a hemoglobin level of 11 g/dl, a hematocrit of 31, and a normal platelet count. Additionally, kidney and liver function tests were normal. An erect abdominal X-ray revealed radioopaque shadows at both the terminal ileum and duodenum (Figure 1).

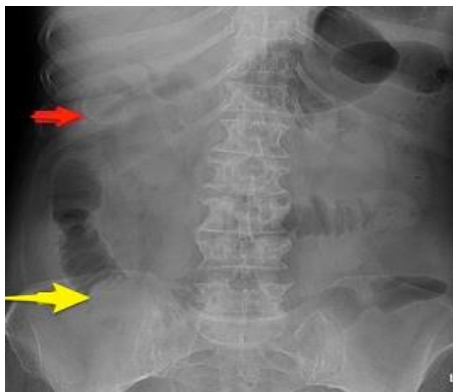


Figure 1: A supine abdominal X-ray reveals the duodenal stone (red arrow) and the ileal stone (yellow arrow).

A computed tomography (CT) of the abdomen revealed dilated jejunal bowel loops with a maximum diameter of 4 cm with a transition zone at the ileum containing peripherally calcified structure (gallbladder stone), in addition to a large stone present at the second part of the duodenum with CEF and pneumobilia (Figures 2 and 3).

Therefore, the diagnosis of synchronous GSI and Bouvert's syndrome was reached. An endoscopic duodenal stone removal was intended, which was unsuccessful due to the stone's size (Figure 4) and non-availability of lithotripsy, then to be followed by surgical ileal stone removal. As a result, the patient underwent exploratory laparotomy with enterotomies for both stones (Figure 5).

However, intra-operatively, the duodenal stone was too large to be pushed into the jejunum, so lateral duodenotomy with stone extraction and subsequent closure transversely in Heineke-Mikulicz fashion was

performed. Two drains were placed, one in the pelvis and the other close to the duodenotomy site. The gall bladder, stomach, small intestine, and large intestine were examined during surgery to ensure that no stone was missed, while the gallbladder and CEF were left with no intervention (due to the patient's age, no stones left in the gall bladder, and two enterotomies).

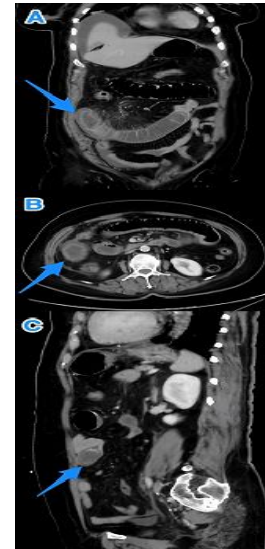


Figure 2: A multiview CT abdomen scan reveals the impacted ileal stone (arrows).

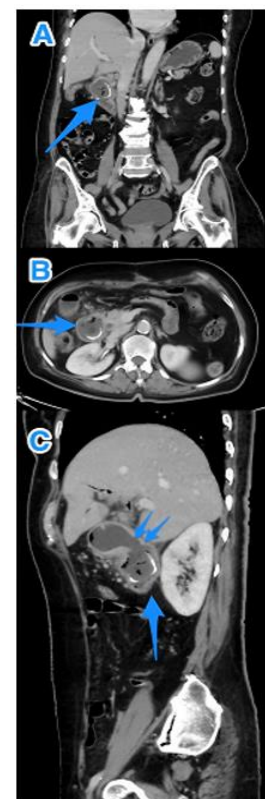


Figure 3: A multiview CT abdomen scan reveals the impacted duodenal stone (large arrows) and the cholecystoduodenal fistula (small arrows).

On the third postoperative day, a CT abdomen with oral contrast revealed no evidence of leakage. A liquid diet was started and progressed as tolerated. The patient was discharged on the seventh postoperative day and had regular follow-ups in the outpatient clinic (1, 6, and 12 weeks) with no complications.



Figure 4: Endoscopic visualization of the duodenal stone.

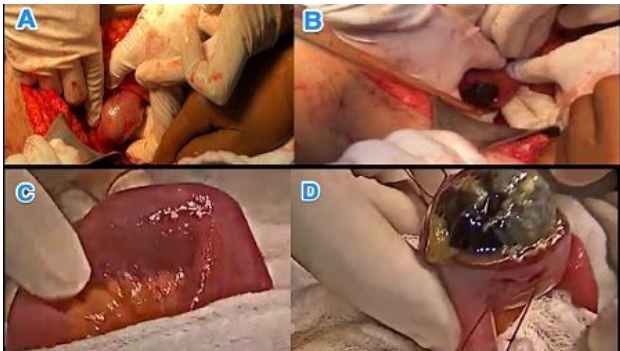


Figure 5: Intra-operative views illustrating the (A and B) duodenal; and (C and D) ileal stone extractions.

DISCUSSION

It is unclear whether complications of acquired CEF are an overlooked entity or a truly rare disease. Given that the majority of affected patients are elderly and have pre-existing comorbidities, such complications are associated with substantial morbidity and mortality, estimated at 60% and 30%, respectively.^{1,3,5} The clinical presentation of GSI differs from that of Bouveret syndrome, whereas the biochemical characteristics, such as leukocytosis and electrolyte disturbance, are unreliable or nonspecific.^{1,3,6,7}

A delay in presentation could be explained by the tumbling phenomenon, which describes the intermittent course of symptoms caused by temporary stone impaction followed by dislodgement of the stone, which then travels distally and re-impacts.^{3,6,8} Similarly to the present case (concomitant GOO and SBO), GSI should be suspected in patients presenting with SBO and having a history of gallbladder stones (a well-known risk factor).⁵ Rigler's triad is comprised of the following imaging findings suggestive of GSI: bowel obstruction, pneumobilia, a

radioopaque shadow within the bowel lumen (ectopic gall stones), and, more recently, a change in gallstone position on serial abdominal X-rays.^{1,3} The abdominal CT scan has become the gold standard for diagnosing GSI (in approximately 60% of cases) and guiding the patient's treatment plan.^{1,3} Magnetic resonance cholangiopancreatography (MRCP) may be advantageous because it can distinguish fluid from stones, as 15-25% of stones are attenuated.^{1,3} Bouveret's syndrome is typically diagnosed by upper gastrointestinal tract endoscopy, as the stone can be detected in 69% of patients.^{3,4} Furthermore, endoscopic findings include compression on the duodenal wall or partially visualized stone, a large amount of stomach contents retained, gastritis, edema, and ulcerations at the site of impact.^{3,5} In both conditions, monitoring vital signs with serial abdominal examinations, gastric decompression through the nasogastric tube, fluid resuscitation, and electrolyte correction must be initiated.⁵

The mainstay of treatment for GSI is surgical intervention (enterolithotomy) with the aim of alleviating bowel obstruction through one or two staged surgeries, which is dependent on the patient's risk stratification.^{9,10} Low-risk patients may undergo biliary intervention concurrently with enterolithotomy, whereas high-risk patients may undergo biliary intervention at a later date or develop recurrences.^{9,10} While endoscopic management with lithotripsy using different modalities is the first-line treatment for Bouveret's syndrome, the optimal approach is based on an algorithm proposed by Khan et al., which focuses on the patient's stability.^{9,11} After medical optimization and patient risk stratification (high risk or low risk), the algorithm provides stepwise management.^{9,11} Therefore, esophagogastroduodenoscopy (EGD) with stone fragmentation is considered for stable patients, while operative enterolithotomy is for unstable patients or in the case of EGD failure.^{9,11} Additional considerations should be given, such as fistula repair in stable and low-risk patients or laparoscopic enterolithotomy following endoscopic lithotripsy if residual stone fragments are substantial.¹¹ Despite the fact that stones up to 3 cm have been successfully removed endoscopically, stones larger than 2.5 cm are more difficult to remove endoscopically, with a failure rate of 78%, necessitating surgical intervention in 42% of patients, which is the situation in the current case.^{1,2,11} However, both conditions have been reported to have a high conversion rate with laparoscopic intervention.¹ Enterotomy for stone retrieval alone or combined enterotomy with cholecystectomy and fistula closure is surgical options for GSI.^{1-3,9} While in Bouveret's syndrome, the most commonly reported techniques for stone removal involve pushing the stone into the jejunum or stomach.^{1-3,9} However, in the present case, the stone was too large to be pushed into the stomach or jejunum; therefore, lateral duodenotomy was performed.

During operative intervention for bowel obstruction in patients with a history of gallstone disease, it is crucial to emphasize the importance of a thorough examination of the gastrointestinal tract (from the stomach to the upper

rectum).^{2,3} Moreover, repair of CEF in patients managed by endoscopic procedures or simple enterotomy is deemed unnecessary due to spontaneous closure, especially in the presence of a patent cystic duct and the absence of residual stones (in the current case, no intervention was made).^{1,3} Furthermore, management should be based on a multidisciplinary approach and tailored to the patient at the discretion of the surgeon and the capabilities of the institution.

CONCLUSION

Rare cholelithiasis complications include GSI, Bouveret's syndrome, and a combination of both. They continue to pose diagnostic and therapeutic difficulties, necessitating an immediate multidisciplinary approach to prevent significant morbidity and mortality. Further reports or larger-scale studies are encouraged to determine the true prevalence and the most effective management approach.

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REFERENCES

1. Alemi F, Seiser N, Ayloo S. Gallstone Disease: Cholecystitis, Mirizzi Syndrome, Bouveret Syndrome, Gallstone Ileus. *Surg Clin North Am*. 2019;99(2):231-244.
2. Liu YY, Bi SY, He QR, Fan Y, Wu SD. Developments in the Diagnosis and Management of Cholecystoenteric Fistula. *J Invest Surg*. 2022;35(11-12):1841-6.
3. Wang F, Du ZQ, Chen YL, Chen TM, Wang Y, Zhou XR. Bouveret syndrome: A case report. *World J Clin Cases*. 2019;7(23):41449.
4. Nuño-Guzmán CM, Marín-Contreras ME, Figueroa-Sánchez M, Corona JL. Gallstone ileus, clinical presentation, diagnostic and treatment approach. *World J Gastrointest Surg*. 2016;8(1):65-76.
5. Hussain J, Alrashed AM, Alkhadher T, Wood S, Behbehani AD, Termos S. Gall stone ileus: Unfamiliar cause of bowel obstruction. Case report and literature review. *Int J Surg Case Rep*. 2018;49:44-50.
6. Abich E, Glotzer D, Murphy E. Gallstone Ileus: An Unlikely Cause of Mechanical Small Bowel Obstruction. *Case Rep Gastroenterol*. 2017;11(2):389-95.
7. Halabi WJ, Kang CY, Ketana N, Lafaro KJ, Nguyen VQ, Stamos MJ, et al. Surgery for gallstone ileus: a nationwide comparison of trends and outcomes. *Ann Surg*. 2014;259(2):329-35.
8. Fearon NM, Kearns EC, Kennedy CA, Conneely JB, Heneghan HM. The impact of ursodeoxycholic acid on gallstone disease after bariatric surgery: a meta-analysis of randomized control trials. *Surg Obes Relat Dis*. 2022;18(1):77-84.
9. Evola G, Caramma S, Caruso G, Dapri G, Evola FR, Reina C, Reina GA. Bouveret's syndrome as a rare complication of cholelithiasis: Disputes in current management and report of two cases. *Int J Surg Case Rep*. 2020;71:315-8.
10. Shelton J, Samad MA, Juhng J, Terry SM. Unusual Presentation of Bouveret Syndrome Resulting in Both Gastric Outlet Obstruction and Small Bowel Obstruction with Perforation. *Medicines (Basel)*. 2022;9(3):24.
11. Khan A, Sanford D, Chapman WC. Treatment strategy formangement of bouveret syndrome. *HPB*. 2017;19:S140.

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