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International Journal of Surgery Case Reports

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Case report

One-stage procedure for the management of Bouveret's syndrome: A case report and operative video

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ARTICLE INFO

Keywords: Bouveret's syndrome Gastric outlet obstruction Enterolithotomy Laser lithotripsy

ABSTRACT

Introduction and importance: Bouveret's syndrome is a rare condition that causes gastric outlet obstruction, due to migrating gallbladder stone (s). It occurs more commonly in elderly female patients with multiple comorbidities and presents with non-specific symptoms. This may lead to delayed definitive management and an increase in morbidity and mortality rate.

Case presentation: We report a case of a 60-year-old female who presented with five days history of upper abdominal pain, nausea and vomiting. She was initially diagnosed as acute cholecystitis, then with further investigation a diagnosis of Bouveret's syndrome was established. An initial attempt at endoscopic stone extraction with mechanical instruments as well as laser lithotripsy was unsuccessful. The patient then underwent a successful one-stage operation for stone extraction and repair of the fistula.

Clinical discussion: There is still a controversy in the decision of treatment of choice. Less invasive options like endoscopy and laser lithotripsy have a low success rate. Therefore, surgical management remains the mainstay of treatment. A one-stage procedure is typically recommended for younger, healthier patients as it reduces the risk of recurrent complications and the need for repeated anesthesia. A two-stage approach may be more appropriate for older, more comorbid patients because of less operative time and lower mortality risks.

Conclusion: Currently, there are no clear management guidelines. Although endoscopic modalities should be considered first, surgical option is the main treatment. We advocate for a one-stage procedure unless the patient is critically ill or if there is no backup hepatopancreaticobiliary surgeon in the healthcare institution.

1. Introduction

Bouveret's syndrome, a rare form of gallstone ileus, is named after French physician, Leon Bouveret, who published two case reports about this syndrome in 1896 [1]. This syndrome is characterized by gastric outlet obstruction (at the pylorus or duodenum), which occurs due to the passage of large gallstone through a fistula tract. Six factors are usually involved in this syndrome. It includes history of cholelithiasis, repeated episodes of cholecystitis, female gender, age greater than 60 years, comorbidities, and gall stones size greater than 2.5 cm in diameter [1,2]. It is a rare syndrome that comprise only 1 % to 3 % of gallstone ileus cases [1]. Currently there is no agreed upon diagnostic approach for detecting this syndrome in patients with symptomatic cholelithiasis.

This may result in a delay in proper management and consequently elevated risk of mortality (12 % to 30 %) [1]. Endoscopic intervention is often attempted first, as it has lower rates of mortality and morbidity (1.6 %) compared to the surgical approach (17.3 %) [1]. However, it has low success rate (43 %), may require multiple sessions, and may result in incomplete stone fragment extraction and subsequent gallstone ileus. [1]. When endoscopic intervention fails, a surgical intervention is indicated. The literature described three main strategies: enterolithotomy alone, enterolithotomy with concurrent cholecystectomy and fistula repair (one-stage procedure), or enterolithotomy with interval cholecystectomy and fistula repair (two-stage procedure) [1]. Here, we present a case that underscores the challenges in reaching this rare diagnosis, as well as the feasibility and success of a one-stage

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https://doi.org/10.1016/j.ijscr.2025.111719

Received 13 March 2025; Received in revised form 9 July 2025; Accepted 21 July 2025 Available online 22 July 2025

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operation.

This case report has been reported in line with the Surgical CAse REport (SCARE) criteria. [3]

2. Case presentation

A 60-year-old female, with past medical history of sickle cell trait, hypertension, bronchial asthma, obesity (Body mass index [BMI]: 53 kg/ m²) and obstructive sleep apnea, presented to the emergency department with five days history of abdominal pain. The pain started suddenly, and was colicky in nature, severe (10/10), located in the epigastric area, radiated to the right shoulder, and alleviated by analgesia. It was associated with nausea and multiple episodes of non-bloody vomiting (food content). She denied fever, chest pain, shortness of breath, changes in bowel habits, jaundice, dark urine, and pale stool. Prior to this episode, she presented to the emergency department twice in the previous week with epigastric pain and vomiting and was managed as gastroenteritis and discharged home. She also had similar pain 2 years prior to this presentation that resolved without interventions. Her surgical history was positive for tonsillectomy, and she had no relevant family, social, or psychiatric history. On Physical examination, she was alert, awake and oriented, and hemodynamically stable (Temperature: 36.9 degrees Celsius, blood pressure: 138/79 mmHg, pulse rate: 65 beats/min, respiratory rate: 22 breath/min, oxygen saturation: 96 % on room air). The abdomen was soft and lax, and not distended, with no rigidity or guarding, however, there was severe epigastric / right upper quadrant tenderness and a positive Murphy's sign. Initial laboratory tests were as follows, with normal reference ranges provided in parentheses: white blood cell count 8.1 $(4-11 \times 10^3)$ μL), C-reactive protein 3.8 (0.1–0.5 mg/dL), erythrocyte sedimentation rate 89 (0-30 mm/h), albumin 3.8 (3.2-5.2 g/dL), total protein 8.2 (6.4-8.3 g/dL), alkaline phosphatase 64 (46-122 U/L), alanine aminotransferase 6 (5-55 U/L), aspartate aminotransferase 12 (5-34 U/L), total bilirubin 0.5 (0.2-1.2 mg/dL), direct bilirubin 0.20 (0.10-0.50 mg/ dL), gamma-glutamyltransferase 23 (9-36 U/L), lactate dehydrogenase 213 (125-220 U/L), amylase 92 (25-125 U/L), and lipase 32 (8-78 U/ L). Biliary ultrasound showed cholelithiasis, gallbladder wall thickening (74 mm), trace pericholecystic fluid, and positive sonographic Murphy's sign. Thus, the patient was admitted as a case of acute calculous cholecystitis and was started on antibiotic therapy. Initially, she experienced improvement in abdominal pain; however, she continued to report gradually worsening nausea, vomiting, and food intolerance despite being on dual antiemetics and discontinuing possible offending

antibiotic (Metronidazole). On the seventh day of hospital stay, the abdominal pain worsened, and she had an episode of bilious vomiting. A contrast-enhanced computed tomography (CT) of the abdomen revealed gallbladder wall thickening with submucosal edema and a fistulous communication with adjacent duodenal segment, as well as pneumobilia. The duodenal wall was thickened with enhancement and surrounding fat stranding. Notably, there was a well-defined intraluminal lesion with heterogenous density and rim calcification, measuring 6.5 \times 4.5 cm, in the fourth part of the duodenum (D4) and causing duodenal D4 outlet obstruction (proximal duodenal dilatation and distal jejunal collapse). This lesion could represent a migrating gallstone; however, malignancy could not be ruled out (Fig. 1). An upper gastrointestinal (GI) endoscopy showed a very large gallstone impacted in the 4th part of the duodenum (Fig. 2a), with failed mechanical extraction using grasper, snare and Roth Net. Another session of upper GI endoscopy was performed under Monitored Anesthesia Care (MAC) and Laser Lithotripsy was attempted for more than three hours but the gallstone did not break, and the procedure was aborted due to time limitation (Fig. 2b / 2c). The patient was then taken for diagnostic laparoscopy which showed extensive pericholecystic adhesions, as well as a small area of suspected duodenal (D4) perforation at the location of the impacted gallstone. Thus, a decision was made to convert the operation to midline laparotomy. Duodenal Kocherization was performed, which identified a cholecystoduodenal fistula at first part of the duodenum (D1). The impacted gallstone was milked proximally to the level of the fistula. The fistula tract was dissected and divided, and the fistula duodenal opening was excised. The fistula opening was further extended longitudinally and a large gallstone ($\sim 7 \times 5$ cm) was extracted through it (Fig. 3). A nasogastric tube was inserted and advanced to the proximal jejunum. Duodenoplasty was then performed (two-layer transverse closure technique) and protected with an omental patch. Methylene blue leak test was negative. The small bowel was run down from the duodenojejunal junction to the cecum and was clear of other gallstones. A Fundus-first cholecystectomy was performed, and abdominal drains were placed. The procedure was completed successfully, in 460 min, with blood loss estimated around 200 mL. Five days after the procedure, the nasogastric tube and all the drains were removed. Histopathology evaluation found extensive cholecystitis and inflammation of the fistula tract with no evidence of malignancy. The patient made an uneventful recovery and was discharged home in postoperative day ten in stable condition.

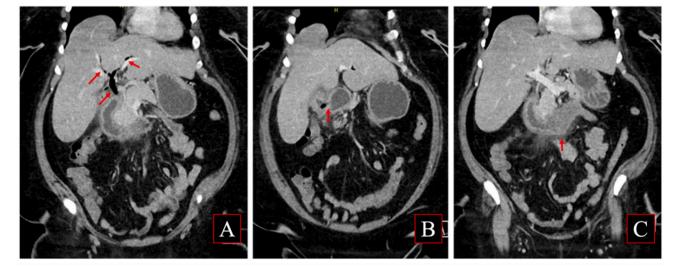


Fig. 1. Contrast-Enhanced CT Scan of the Abdomen and Pelvis Figure legends: (A) Pneumobilia, (B) Cholecystoduodenal fistula, (C) Intraluminal duodenal impacted gallstone.

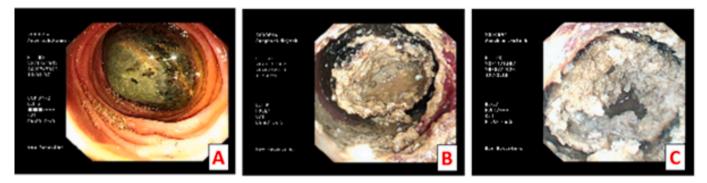


Fig. 2. Upper Gastrointestinal Endoscopy with Trial of Endoscopic Laser Lithotripsy Figure Legends: (A) Impacted gallstone in fourth part of the duodenum, (B, C) Gallstone after attempted laser lithotripsy.



Fig. 3. Shape and Size of Gallstones found in the duodenum and gallbladder (Units of measurement is millimetre; mm).

3. Discussion

Bouveret's syndrome remains a challenging diagnosis to make due to rarity and unavailability of clear guidelines. We report a case of a 60-year-old multimorbid female that was managed successfully with a one-stage procedure. Our patient was initially admitted with grade II (moderate) acute calculous cholecystitis, based on the Tokyo Guidelines (symptom duration >72 h), and had an American Society of Anesthesiologists Physical Status (ASA-PS) classification of III (morbid obesity; BMI: 53 kg/m^2). Thus, she was considered high-risk for early surgical intervention, and the decision was made to pursue conservative management, with a plan for delayed—rather than the usual early—cholecystectomy, which is typically reserved for mild disease in low-risk patients [4].

As underlined in the literature, reaching a timely diagnosis of Bouveret's syndrome is challenging due to the variability of its presentation. This syndrome may have acute, subacute, or chronic symptoms, and usually presents with non-specific GI symptoms, with or without initial evidence of GI obstruction [5]. Thus, it has a wide differential diagnosis and several disease mimickers. The diagnostic challenge may be compounded if it presents concurrently with acute cholecystitis (as in our case), or with other acute or chronic diseases (e.g., peptic ulcer disease, cholangitis, malignancy). In addition to the above, other factors may have contributed to the delay in performing further diagnostic workup and reaching the final diagnosis in our case. First, the patient did not present with clear symptoms of bowel obstruction—there was no bilious or projectile vomiting, no abdominal distension (unlike in gallstone ileus), and no obstipation or changes in bowel habits. Second, utilizing biliary ultrasound as the initial imaging modality, although arguably a

sound decision, may have introduced imaging bias, steering us toward the more common diagnosis of acute cholecystitis. Third, there was initial clinical improvement, reassuring vital signs, normal liver enzymes and electrolytes, as well as a downward trend in inflammatory markers (CRP). Finally, the initial choice of antibiotics may have played a part as well. Our institutional antibiogram recommends the use of ciprofloxacin or ceftriaxone, plus metronidazole, as the first-choice empirical regimen for mild to moderate acute cholecystitis. The latter, however, is a well-known drug that may induce nausea and vomiting in some patients [6]. After stopping this medication, we waited for a couple of days to observe her clinical progression. Our case presentation underscores these challenges clearly. Despite working in a high-resource setting, with direct access to advanced imaging modalities and diagnostic/therapeutic endoscopy, the aforementioned challenges precluded us from obtaining a CT scan until hospital day seven, by which point the final diagnosis of Bouveret's syndrome was made. Currently, there are no clear guidelines for the diagnostic approach to Bouveret's syndrome. Physicians should maintain a high index of suspicion, particularly in elderly patients with protracted symptoms, and adopt a low threshold for utilizing advanced imaging.

Our case initially underwent less invasive management with endoscopy to extract the gallstone, followed by an attempt to fragment it using laser lithotripsy which both ended up unsuccessful [5,7]. This is expected as the success rate of endoscopic management is not very high [7]. Another possible option is extracorporeal shock wave lithotripsy which also has a low success rate. It is essential to keep in mind that these techniques although less invasive carry risk of complications including bleeding, perforation, and gallstone ileus especially if the gallstone was fragmented as parts may dislodge into the terminal ilium

causing obstruction that is not reachable by endoscopy [8]. Even in the case of successful management with less invasive techniques, a definitive surgical management should be considered because there is risk of recurrence with other stones passing as the rate of spontaneous fistula closure is only 50 %. In fact, 91 % of patients will require definitive surgical management [7]. Also, other gallstones distal to this obstruction may be missed emphasizing the importance of examining the entire gastrointestinal tract during surgery. [9]

Surgical management remains the mainstay of treatment, however, there is still controversy in regards to the most appropriate surgical operation [10] [7]. Currently the literature has mentioned three main methods including a enterolithotomy alone to alleviate the obstruction, a one-stage procedure (enterolithotomy with cholecystectomy and fistula repair) or a two-stage procedure (Initial enterolithotomy followed by interval cholecystectomy and fistula repair) [7]. Regardless of whether a one- or two- stage procedure is attempted, a less invasive techniques with laparoscopy or robotic surgery should be attempted especially when an experienced surgeon is available [7,10]. The literature generally suggests that patient age, comorbidities, and patient general condition are the deciding factors to the surgical approach. A one-stage procedure is generally recommended in younger healthier patients while a two-stage procedure in older multi-comorbid patients [7,11]. A one-stage procedure has the advantage of reducing the risk of repeated general anesthesia and preventing recurrence of future complications, including acute cholecystitis, acute cholangitis, gallstone

Table 1Therapeutic Options for Bouveret's Syndrome.

Treatment Option	Description	Advantages	Disadvantages
Endoscopic Extraction *	Stone removal using mechanical via upper endoscopy (Roth net retrieval)	Minimally invasive; lower morbidity/ mortality	Low success rate; may require multiple sessions; risk of incomplete extraction, perforation, bleeding
Laser Lithotripsy	Endoscopic fragmentation of the stone using laser via upper endoscopy	Useful for hard stones that cannot be extracted whole	Time-consuming; may fail; risk of incomplete fragmentation or gallstone ileus, perforation, bleeding
Extracorporeal Shock Wave Lithotripsy (ESWL)	Non-invasive fragmentation of stones using external shock waves	Non-invasive	Low success rate; not widely available
Enterolithotomy Alone	Surgical removal of the stone without addressing the fistula or gallbladder	Shorter operation time; less invasive; lower mortality risk	Risk of recurrent symptoms; residual gallstones; fistula- related complications, risk obscured malignancy
One-stage Procedure	Enterolithotomy + cholecystectomy + fistula repair (all in one surgery)	Definitive treatment; prevents recurrence; avoids reoperation and repeat anesthesia	Longer operative time; higher complexity; risk of bile duct injury
Two-stage Procedure	Enterolithotomy first; delayed cholecystectomy and fistula repair in a second operation	Shorter initial surgery; suitable for critically ill or unstable patients	Requires second surgery; possible recurrence or complications during interval period

 $^{^{*}}$ Other endoscopic options include: Electrohydraulic lithotripsy, and triangle tip (TT) electrosurgical knife. Both are not widely available.

ileus, and gallbladder cancer. On the other hand, a two-stage approach is associated with less operative time and lower risk of mortality (Table 1) [7,9].

Our case underwent a one-stage procedure with entreolithotomy through the fistula opening site at D1 and proximally milking the stone. This was done, taking into account the friability of the distal duodenal wall, to reduce the risk of perforation when distally milking the stone into the jejunum. This was followed by closure of the fistula with placement of omental patch and finally a cholecystectomy. Although literature is contradictory regarding whether to do cholecystectomy in patients with gallstone ileus, a newly published study in 2023 found no statistical difference in risk of surgical site infection, 30-day mortality, and sepsis among patients who underwent enterolithotomy with concurrent cholecystectomy as compared to enterolithotomy alone. [12]. Taking into consideration that the presence of multiple gallstones within the gallbladder in Bouveret's syndrome, as we have demonstrated in our case, would place these patients at an increased risk for future complicated cholelithiasis. Thus, in this situation, a cholecystectomy is indicated to decrease risk of complications [7]. Its important to note, that a concurrent cholecystectomy, place the patient at risk of bile duct injury during dissection due to the severe inflammation and possible anatomical anomalies [13]. Therefore, we only advocate a one-stage procedure in patients who are not critically ill or in severe sepsis / septic shock, or have retained gallbladder stones, in the presence of an experienced, preferably a hepatopancreaticobiliary, surgeon. Otherwise, an enterolithotomy alone, or a two-stage procedure will be more appropriate.

4. Conclusion

Bouveret's syndrome is a rare condition that presents with non-specific symptoms and can mimic common upper abdominal pathologies. Therefore, a high index of suspicion, along with the use of diagnostic modalities such as computed tomography and endoscopy, is essential to establish the diagnosis. Currently, there are no clear management guidelines, however, endoscopic modalities should be considered as first line. Surgical option is the main treatment in case of failed endoscopic management. We advocate for a one-stage procedure unless the patient is critically ill or if there is no backup hepatopancreaticobiliary surgeon in the healthcare institution.

Consent

Written informed consent was obtained from the patient for publication and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Ethical approval

Case reports do not require ethical approval in our institution.

Guarantor

Husain Ali Alsaffar.

Funding

This case report is not funded.

Author contribution

Husain Nader Alshaikh: Review and editing.
Shadi A. Alshammary: Review and editing.
Husain Ali Alsaffar: Discussion and conclusion.
Ahmed osama almubayyed Literature review and conclusion.
Razan Saud Al Nimer: Introduction and case description.
Sajedh Najeh Al kazim Introduction and case description.

Conflict of interest statement

The authors declared no conflicts of interest with respect to the authorship and/or publication of this article.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.ijscr.2025.111719.

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