



Case report

Concomitant Bouveret's syndrome and biliary obstruction: A tailored treatment approach for an elderly patient

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ABSTRACT

Introduction and importance: Bouveret's syndrome is a very rare form of gallstone ileus, that occurs when a sizable gallstone enters the gastrointestinal tract via a bilioenteric fistula and is impacted in the pylorus or proximal duodenum, causing gastric outlet obstruction. It usually occurs in a geriatric population with multiple comorbidities, and causes significant morbidities and mortalities.

Case presentation: An 88-year-old patient with concomitant Bouveret's syndrome and biliary obstruction was presented. The duodenal obstructed stone and biliary stone were successfully removed by endoscopic approach. The patient resumed diet on day-1 and recovered smoothly.

Clinical discussion: Due to the rarity of Bouveret's syndrome, there are no standardized recommendations for the management of these patients, including open, laparoscopic surgical approach or endoscopic approach. Minimally invasive treatment was tailored to the condition of the patient and clinical findings.

Conclusion: The best approach is the one tailored to each patient, with the consideration of the patient's medical condition, age, comorbidities, life expectancy, and available expertise. This article highlights the key features of the disease, and the precautions during endoscopic treatment.

1. Introduction

Bouveret's syndrome is characterized by the passage of a large gallstone through a cholecysto-gastric or cholecysto-duodenal fistula in the stomach or duodenum resulting in gastric outlet obstruction. The first case of duodenal obstruction by a gallstone was described by Beaussier in 1770. Bonnet reported two similar cases discovered during autopsy in 1841.

In 1896, the disease acquired the name of Bouveret's syndrome after the French physician Leon Bouveret, who published 2 case reports in *Revue de Medecin* [1]. The biliary-enteric fistula is formed because of chronic cholecystitis, resulting in the inflammation and adhesion of the gallbladder to the adjacent wall of gastrointestinal tract, together with mechanical pressure due to gallstones impaction in the gallbladder itself. This results in pressure necrosis of the apposed gallbladder and enteric wall, and the gallstones eroded into the adjacent gastrointestinal tract through the fistula tract. Bouveret's syndrome's prevalence is highest among elderly women with >2.5 cm gallstone [2,3]. Early diagnosis is difficult because of its rarity and the absence of typical symptoms. Treatment consists of endoscopic or surgical removal of

obstructive stone. Concomitant or interval cholecystectomy and excision of the cholecysto-enteric fistula may be needed. Additionally, an elderly age group with significant comorbidities is usually afflicted, so the choice of treatment needs to be appreciative of the condition of the patient and optimized for optimal recovery. Due to the rarity of Bouveret's syndrome, there are no standardized recommendations for the management of these patients, including open, laparoscopic surgical approach or endoscopic approach.

An 88-year-old patient with concomitant Bouveret's syndrome and biliary obstruction was presented. Minimally invasive treatment was tailored to the condition of the patient and clinical findings.

This case report has been reported in line with the SCARE 2020 criteria [4].

2. Case presentation

An 88-year-old female was admitted to our hospital via Accident and Emergency Department with 3 days of epigastric pain and a day of profuse postprandial vomiting. She had no symptom of gastrointestinal bleeding. The patient had a known history of hypertension, type II

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diabetes mellitus, duodenal ulcer, and gallstones. On physical examination, there were no signs of jaundice, abdominal distension, or peritonitis. Initial blood tests showed normal liver function. White cell count was elevated to $14 \times 10^9/L$, and hemoglobin was 9 g/dL. On investigation, an abdominal X-ray was first performed which showed signs of pneumobilia in the right upper quadrant, but there were no dilated bowel loops (Fig. 1). Urgent contrast enhanced Computed Tomography (CT) scan of the abdomen was suggestive of chronic cholecystitis with gallstones, a dropped stone in the common bile duct (CBD) that caused biliary obstruction, a possible cholecysto-duodenal fistula, as well as an impacted gallstone at the duodenum that caused gastric outlet obstruction (Fig. 2). Occurrence of Bouveret's syndrome was evident at this point and the diagnosis was established. An endoscopic treatment was chosen due to the patient's advanced age. A one-stage endoscopic approach for removal of stone in duodenum and biliary decompression was planned. Oesophago-gastro-duodenoscopy (OGD) and endoscopic retrograde cholangiopancreatography (ERCP) was performed by senior surgeons under general anesthesia at day 1 after admission. OGD showed a 3 cm stone impacted at 1st part of duodenum (D1) and 1st/2nd junction of duodenum (D1/2) (Fig. 3). At first, electrohydraulic lithotripsy (EHL) under saline irrigation was used to fragment the impacted stone in duodenum. The stone fragments were flushed down to the following gastrointestinal tract and the endoscope was able to pass down to the D2 with the Ampulla of Vater seen. The fistula was suspected to be at the D1/D2 region and the area was mildly edematous. ERCP was performed then. The CBD was cannulated and a limited cholangiogram showed dilation of the biliary tree with a 1 cm biliary stone inside the lower common hepatic duct (Fig. 4). Due to the patient's advanced age, acute condition and avoiding prolong general anesthesia, we have not tried to remove CBD stone in the same session. Instead, a sized 10 French, 8 cm long plastic biliary stent was inserted into the CBD for biliary drainage first. A nasogastric tube was then inserted over guide-wire under fluoroscopic guidance at the end of the procedure for bowel decompression. The patient was monitored closely after the procedure and oral diet was resumed on post-procedure day 1. Bile culture showed *Enterococcus faecalis*, *Escherichia coli*, *Clostridium perfringens*, *Candida albicans*. A course of antibiotic was completed also. She was discharged



Fig. 1. Abdominal X-ray showed signs of pneumobilia (annotated with arrow) in the right upper quadrant, but there were no dilated bowel loops.



Fig. 2. Abdominal CT scan showing a calcified stone impacted in the first portion of duodenum.

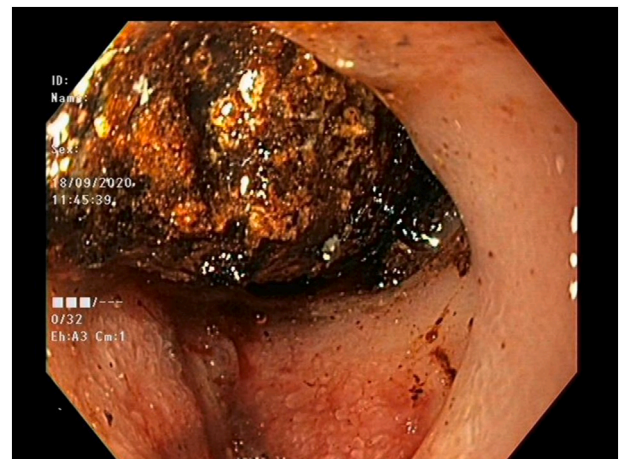


Fig. 3. OGD showed a 3 cm stone impacted at 1st part of duodenum (D1) and 1st/2nd junction of duodenum (D1/2).

from the hospital on post-procedure day 5. One month later, a follow-up ERCP was performed to remove the CBD stone. The patient was well with good oral intake. During follow-up at 2-year after the procedure, she had no gallstones related complications, and she was well.

3. Discussion

Bouveret's syndrome represents only 1–3 % of all gallstone related mechanical obstructions in the gastrointestinal tract, which by itself only constitutes about 1–4 % of all small bowel obstructions. The rarity of the disease can be explained as only 0.3–0.5 % of gallstones develop biliary-enteric fistulas, and even if they do, it depends on the size of the gallstone relative to the bowel luminal diameter, most of the gallstones are can either pass uneventfully to be excreted by the patient or with gastrointestinal tract impaction [5]. Majority of these passed gallstones impact in the narrowest part of terminal ileum, and impaction in the pylorus or duodenum is much less common. There is an increased risk of developing Bouveret's syndrome if one has developed gallstones larger than 2.5 cm, which makes the gallstone easier to be incarcerated at the

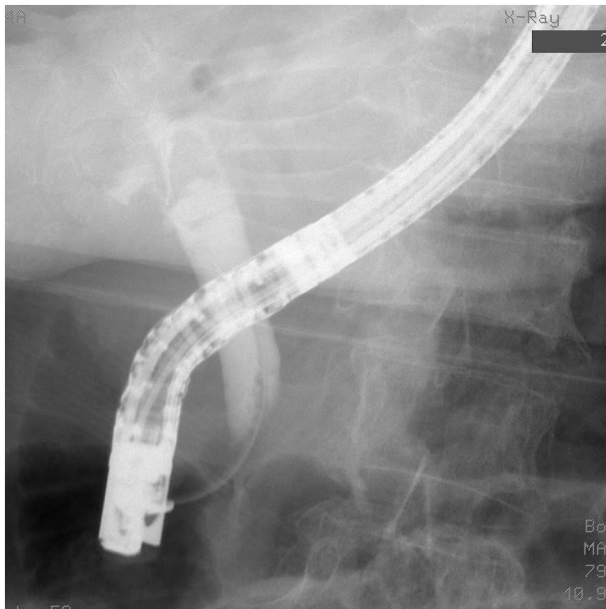


Fig. 4. Cholangiogram showed dilation of biliary tree with a 1 cm biliary stone inside the lower common hepatic duct.

duodenum after its migration through the cholecysto-duodenal fistula. Patients with post-surgical altered gastrointestinal tract anatomy also have an increased risk.

Due to the rarity of Bouveret's syndrome, there is a low awareness of the disease in the differential diagnoses, and there are no standardized recommendations for the management of these patients, including open, laparoscopic surgical approach or endoscopic approach. Bouveret's syndrome usually occurs in an older population who will likely be poor surgical candidates in the presence of multiple comorbidities that can lead to operative complications. Therefore, there is a tendency of delay in diagnosis, and a high morbidity and mortality rate. Therefore, it is essential to increase awareness of its occurrence and establish strategies for diagnosis and treatment. However, Bouveret's syndrome remains difficult to diagnose as the symptoms are nonspecific and the physical examination findings are subtle. Bouveret's syndrome presents with non-specific symptoms, most commonly a triad of epigastric pain, nausea, and vomiting [2]. The differential diagnoses can be various pancreatic, gastric, small bowel, biliary or liver pathologies. Most of the time, the baseline blood test has no significant abnormality. The use of imaging studies in combination with the clinical presentation is important for early diagnosis of Bouveret's syndrome. The classic radiologic sign of gallstone ileus is the Rigler triad (also called Rigler sign): pneumobilia, intestinal obstruction and an ectopic gallstone. However, in Bouveret's syndrome, due to the proximal gastrointestinal tract obstruction, the features of intestinal obstruction such as abdominal distension sign, high-pitched bowel sounds, dilated bowel loops in X-ray etc., may not be present. In most cases, CT scan is needed for diagnosis of Bouveret's syndrome. A CT scan can provide important information about the presence of a biliary-enteric fistula, the level of gastrointestinal tract obstruction, the inflammatory status of the gallbladder and involved gastrointestinal tract, the size, number and locations of the impacted gallstones, and any other concomitant gallstones related complications. Since 15–25 % of gallstones are isoattenuating, it may not be well visualized in CT scan sometimes. OGD may facilitate the diagnosis by showing gastro-duodenal obstruction, the obstructing gallstone or the fistulous opening. However, during the OGD, only approximately 2/3 of gallstones can be visualized and the precautions of prevent aspiration need to be taken [2].

Surgical and endoscopic approaches are the treatment options, with 94 % and 43 % success rate respectively [6]. Surgical options include

enterotomy (gastrotomy or duodenotomy) and stone extraction alone, concomitant enterotomy and stone extraction with cholecystectomy and excision of the cholecysto-enteric fistula (one-stage procedure) or enterotomy and stone extraction with interval cholecystectomy and excision of the cholecysto-enteric fistula after 4–6 weeks (two-stage procedure). Simultaneous cholecystectomy and fistula repair or not, one-stage or two-stage approach of surgery are still debatable [7,8]. Simultaneous cholecystectomy and cholecysto-enteric fistula tract excision can prevent subsequent biliary complications of gallstone related complications. On the other hand, the difficult cholecystectomy, and excision of the large and chronic cholecysto-enteric fistula tract associated with high morbidity and mortality risks, bile duct injury rate, bleeding risk, and the healing problem of the repaired part of duodenum. Also, the healing of such large repaired duodenal fistula opening has potential risk of duodenal stricture. Management of Bouveret's syndrome includes surgical or endoscopic option. Surgery can be performed by open or laparoscopic approach. A major disadvantage of open surgery is that the potential risks of laparotomy wound, enterotomy wound over the duodenum and long general anesthesia for an elderly patient with multiple comorbidities and advanced age. There may be a risk of the healing problem of the repaired duodenal wall, or even worse, leakage may occur, and the gastric juices, saliva, bile present in the duodenum can leak to the peritoneum, causing peritonitis. There is also a risk of stricture of the duodenal lumen after healing, and this can lead to gastric outlet obstruction in the future. Laparoscopic approach is technically demanding, especially the difficult dissection due to chronic adhesions and the suturing of the enterotomy site, but it has the advantage of earlier recovery and less pain.

Endoscopic approach has the advantages of minimally invasive approach such as earlier recovery and less pain, however its success depends on the presence of adequate endoscopic skills [9]. Fragmentation of the obstructed stone by EHL or laser lithotripsy is more feasible and safer for removal of large sized obstructed stone. Other endoscopic procedures such as extraction by endoscopic graspers, nets, baskets, and snares is another option, but the success rate is very low due to insufficient space adjacent to the impacted large stone for the opening of grasping instruments [5,6,10]. There is also a potential risk of esophageal obstruction or injury during extraction of the stone. An esophageal overtube is usually needed to avoid esophageal injury during this endoscopic extraction. During endoscopic treatment, apart from general anesthesia, sedation with monitored anesthesia care (MAC) is another option. This can minimize the anesthetic risk for older patients with potentially higher cardio-respiratory complications. Due to the absence of enterotomy repaired site, patients can resume oral diet earlier and there is no risk of leakage or stricture of the repaired duodenal wall. In our patient, in view of her advanced age, and physical condition, we adopted the endoscopic approach for the obstructed stone removal in duodenum. A few cautions should be paid during endoscopic treatment. 1) Since fragmentation of a large impacted gallstone and saline irrigation are needed, frequent suction of fluid in stomach during procedure to prevent aspiration is very important. 2) Precise application of EHL to the obstructed gallstones is mandatory in order to avoid the risks of bleeding or perforating the duodenal wall due to the mis-application of shock-wave to duodenal wall. 3) the obstructed stone needs to be fragmented into as small as possible, in order to avoid the its migration along the gastrointestinal tract and cause distal post-procedure gallstone ileus in the terminal ileum. 4) In addition, choosing a long enough and adequate diameter of the wire of EHL passing the working channel of OGD scope is essential. After the endoscopic removal of obstructed stone in duodenum, we adopted a conservative approach to the gallbladder and cholecysto-duodenal fistula tract for our 88-year-old patient. Recurrence of large sized gallstones, especially stones of sizes larger than 2 cm (like in cases of Bouveret's syndrome where the gallstone is large enough to cause gastric outlet obstruction), take a long time to form. There is a possibility that in case of a patent cystic duct and the absence of residual sizable stones in the gallbladder, the fistula can close spontaneously. In

our patient, she had no gallstone related complication at 2-year after procedure.

4. Take home messages

In conclusion, a patient with concomitant Bouveret's syndrome and biliary obstruction avoided major surgery due to successful treatment with endoscopic approach. The patient had the advantages of an earlier resumption of oral diet, much less post-procedure pain, much less complication risks and better recovery. The best approach is the one tailored to each patient, with the consideration of the patient's medical condition, age, comorbidities, life expectancy, and available expertise.

Consent

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

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All authors contributed in writing the paper.

Declaration of competing interest

The authors state that they have no conflicts of interest for this report.

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