

Bouveret's syndrome: A rare presentation of gastric outlet obstruction

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Abstract. Bouveret's syndrome refers to gastric outlet obstruction caused by the formation of a cholecystoduodenal fistula with subsequent migration and impaction of a large gallstone into the duodenum. A case of a 59-year-old male who presented to our institution with consistent abdominal pain and nausea is reported herein. Bouveret's syndrome was diagnosed after conducting a computed tomography scan. Surgery was performed wherein gallstone removal was followed by cholecystectomy and fistula repair associated with a pyloric bypass via gastro-jejunostomy. The patient recovered well following surgery and has remained free of symptoms for the last year.

Introduction

Bouveret's syndrome is defined as gastric outlet obstruction (GOO) caused by the duodenal impaction of a gallstone after entering the small bowel through a fistulous tract (1,2). Léon Bouveret initially described this syndrome in 1896, wherein two cases of gastric outlet obstruction due to gall stones were reported (3). As the symptoms are frequently non-specific and initial presentations are often benign, cases of Bouveret's syndrome are at risk of being undiagnosed (4-6). The aim of the present case report, accompanied by a brief overview of current diagnostic and therapeutic modalities, is to enhance awareness of this overlooked clinical entity in order to ensure timely diagnoses and treatments for future patients. This enhanced awareness is also essential for improving the prognosis of this syndrome.

Case report

In the present study, the patient was a 59-year-old male with a medical history of gallbladder stones and an unremarkable surgical history, who presented with complaints of abdominal pain. The pain was described as being located in the mid-upper quadrant of the abdomen and as severe, constant and non-radiating. The pain for the preceding 5 days had also been associated with nausea and non-bilious, non-bloody vomiting. The patient denied any fever, chills, diarrhea, hematemesis, pruritis or melena. On physical examination, the patient's vital signs were within the normal limits, and no masses were palpable. Their abdomen was soft and non-tender. A nasogastric tube was placed, and it drained >1 l fluid. Routine laboratory tests revealed a white blood cell count of $18.4 \times 10^9/l$ (normal range, $4.0-11.0 \times 10^9/l$), an alkaline phosphatase level of 131 U/l (normal range, 40-120 U/l) and a γ -glutamyl transferase level of 116 U/l (normal, <37 U/l) (7). The remaining laboratory test results were unremarkable. The patient underwent a computed tomography (CT) scan, which revealed a gastric outlet obstruction secondary to an impacted gallstone within the duodenum, a cholecystoduodenal fistula and a collapsed gallbladder, with thickening of the gallbladder wall and air present within the gallbladder (Figs. 1 and 2).

An exploratory laparotomy was planned with a working diagnosis of GOO. Intraoperatively, it was observed that the stone was impacted between the first and second part of the duodenum, and that it could not be milked neither proximally nor distally. Then, a Kocher incision was performed. The gallbladder was located tightly adherent to the wall of the duodenum and blunt dissection exposed the two openings of the fistula between the gallbladder corpus and the first portion of the duodenum. A formal removal of the gallbladder was then performed, with simultaneous resection of the fistula. To aid the removal of the gallstone, a duodenotomy that elongated the opening in the duodenal wall was performed (Fig. 3). In order to avoid missing the possible presence of other stones, the entire gastrointestinal tract was then palpated. With one layer of interrupted 2-0 monofilament absorbable sutures, the defect in the duodenal wall was closed and then reinforced with the use of a surgical adhesive material. Finally, a gastrojejunostomy was carried out. One duodenostomy tube was placed inside the duodenum. The size of the extracted

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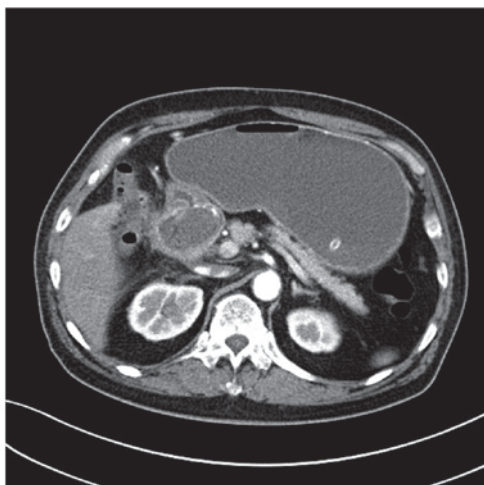


Figure 1. Transaxial contrast-enhanced computed tomography.

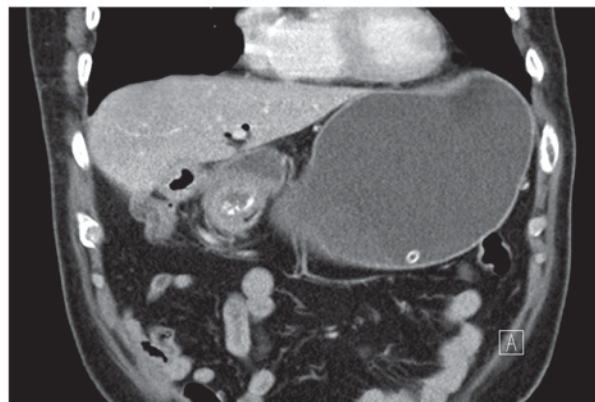


Figure 2. Coronal contrast-enhanced computed tomography.

stone was 6.0x3.5 cm (Fig. 4). The type of gallstone was not assessed, which was a limitation of the present report. The pathological results suggested chronic inflammation of the gallbladder, but the histopathological features were not assessed, which was a further limitation of the present report. The patient recovered well following surgery with no complications and was discharged on the 7th day. The patient is currently on a regular follow up program. The patient had an uneventful recovery and remains in good health at the time of writing.

Discussion

Bouveret's syndrome is an uncommon form of gallstone-ileus caused by large gallstones that reach the duodenal bulb and get lodged there through a biliodigestive fistula (5). The gallstone volume may become enlarged due to an accumulation of fecal matter and salts on its surface (8). Recurrent inflammation of the gallbladder frequently causes erosion and necrosis of the gallbladder wall and allows the formation of biliary-enteric fistulas (9). The size of the stones associated with Bouveret's syndrome are typically >2.5 cm, and it is accepted that larger stones are associated with more proximal obstruction (10).

Risk factors for Bouveret's syndrome include gallstones >2.5 cm in size, the female sex and age >70 years (11). The cluster of symptoms, including abdominal pain, nausea/vomiting, and dyspepsia, is caused by gastric outlet obstruction. Abdominal imaging of gallstone ileus typically reveals Rigler's triad of ectopic gallstone, pneumobilia and small bowel obstruction (12). However, only ~33% of gallstone ileus cases exhibit these changes on conventional radiographs (13). Due to the gastric outlet obstruction, a dilated stomach is expected to be observed on plain abdominal radiographs in cases of Bouveret's syndrome (14). Compared with other imaging methods, ultrasound (US) is more sensitive with respect to detecting ectopic gallstones and pneumobilia. The combination of US and abdominal films has increased the sensitivity to 74% for the diagnosis of gallstone ileus (15).

The Riglar's triad of findings on CT scans is more readily apparent. CT scans can also reveal the presence of a fistula;

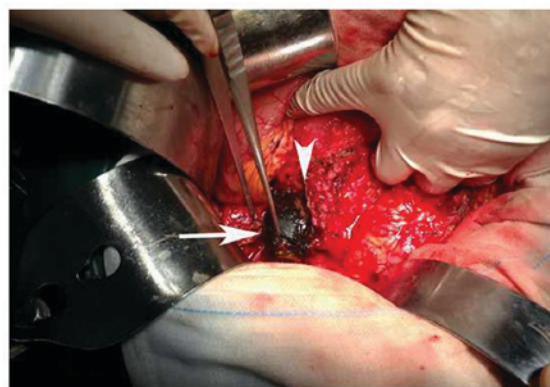


Figure 3. Gall stone in the opened duodenum and cholecystoduodenal fistula. Cholecystoduodenal fistula (arrow head) and stone (arrow).



Figure 4. The extracted ectopic gallstone.

the degree of bowel obstruction; the degree of inflammation in the surrounding tissue; and the size, number and locations of the occluding gallstones. Edema and ischemia of the affected gastrointestinal tract site can be detected by contrast-enhanced CT scans (16,17). Contrast-enhanced CT is of particular importance to making decisions regarding the management of possible bowel ischemia. To help provide correct diagnoses and rule out the presence of intraductal concretions, magnetic resonance cholangiopancreatography (MRCP) can be used.

However, it may be difficult to differentiate concrements and air using MRCP. MR imaging is also sensitive for identifying a fistula and can be used for the confirmation of findings prior to treatment (11,18,19). Esophagogastroduodenoscopy (EGD) allows the visualization of a dilated stomach and the impacted stone, which appears as a hard and non-fleshy mass. It can also reveal the duodenal ostium of the biliodigestive fistula (10). EGD in Bouveret's syndrome is important due to it being of both diagnostic and therapeutic significance.

The primary aim in treating Bouveret's syndrome is to remove the obstructing gallstone. Both nonsurgical (endoscopic) and surgical (open or laparoscopic) approaches are therapeutic options (20). The documented cases of Bouveret's syndrome being treated successfully with endoscopic retrieval are few to date. One such literature review reported that the success rate of endoscopic retrieval alone was only 10% (21). Although endoscopic modalities are less invasive and safer for patients with comorbidities, they carry the risk of serious complications, such as stone having the lodged in the patient's esophagus and fragments lodged in the terminal ileum (22). Therefore, surgery is the main treatment modality for Bouveret's syndrome, especially when percutaneous or endoscopic approaches are not first choice or have failed (23).

The therapeutic strategy should take into account many parameters, including the general condition of the patient, any comorbidities, age, the inflammatory status of surrounding tissues, the location of the obstruction, the size of the fistula and calculus (24), and the number of gallstones. Whether biliary surgery should be carried out at the same time as the relief of obstruction of the bowel (one-stage procedure), performed later (two-stage procedure) or not at all has been a long-standing controversy in the management of gallstone ileus (25,26).

Compared with simple enterolithotomy alone, the one-stage procedure has been associated with higher mortality and morbidity rates. However, considering the appropriate available surgical expertise, low-risk patients should be offered the one-stage definite procedure (27).

In the present case, an open enterolithotomy with cholecystectomy and fistula repair was performed. Gastrojejunostomy was also conducted due to the extensive degree of obstruction in the patient. The noted bulbar ulcer of duodenum may have led to stricture formation. A pyloric bypass via gastrojejunostomy was performed in order to spare the patient from future surgery for GOO (due to a different cause), which would significantly increase their morbidity. Many previous studies have recommended delayed cholecystectomy and fistula repair considering that immediate cholecystectomy or fistula dissection can be associated with significant morbidity and mortality (28,29). As the patient was relatively young with no comorbidities in the present report, an enterolithotomy with cholecystectomy and fistula repair was performed successfully. The patient had an uneventful recovery and remains in good health at the time of writing.

In conclusion, Bouveret's syndrome is a rare form of gallstone ileus. A tailored surgical strategy considering the patient's age, general and local inflammatory status, and the presence of comorbidities, in association with the morbidity and mortality rates of each method should be applied for the successful management of Bouveret's syndrome.

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Availability of data and materials

All data generated or analyzed during this study are included in this published article.

Authors' contribution

Y-BY, YS, J-BX and F-ZQ contributed to the conception, design and data interpretation. All authors read and approved the manuscript.

Ethics approval and consent to participate

This report was approved by the Institutional Review Board and Human Ethics Committee of the Affiliated Huaian No. 1 People's Hospital of Nanjing Medical University (Huaian, China). The patient provided written informed consent to participate.

Patient consent for publication

Consent for publication was obtained from the patients.

Competing interests

The authors declare that they have no competing interests.

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