

Endoscopic therapy for Bouveret syndrome, illustrated by a case report

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Abstract

Bouveret syndrome is an exceptionally rare form of gallstone ileus secondary to a bilioenteric fistula, through which a voluminous gallstone can migrate into the pylorus or duodenum, thereby causing gastric outlet obstruction. In order to increase awareness, we reviewed the clinical features, diagnostic tools and management options for this uncommon entity. We specifically focus on endoscopic therapeutic options, illustrated by a case of a 73 year old woman with Bouveret syndrome, where endoscopic electrohydraulic lithotripsy was successful in relieving gastroduodenal obstruction. (Acta gastroenterol. belg., 2023, 86, 360-362).

Keywords: Bouveret, gallstone ileus, endoscopy, lithotripsy.

Introduction

Bouveret syndrome was named after the French physician Léon Bouveret, who published 2 case reports in 1896 of gastric outlet obstruction caused by gallstones (1,2,3). It describes the migration of a stone from the gallbladder to the bowel through a bilioenteric fistula caused by chronic inflammation of the gallbladder, resulting in gastric outlet obstruction secondary to an impacted gallstone (1).

Bouveret syndrome therefore is a rare form of gallstone ileus, comprising only 2-3% of intestinal obstructions caused by gallstones (1). The site of occlusion in gallstone ileus usually is the most narrow caliber of the distal intestine, being the terminal ileum (3). Bouveret syndrome however, describes gastric outlet obstruction due to a more proximally impacted gallstone.

Considering that Bouveret is a complication of cholelithiasis, its prevalence is also highest among elderly women, with a median age at presentation of 74 years and a female to male ratio of 1.9 (1,3).

Despite the rarity of the syndrome, it is accompanied with significant morbidity and mortality, so awareness for the syndrome and the possible therapeutic strategies should be raised.

Case history

A 73 year old woman, in treatment for metastatic renal cell carcinoma (RCC), presented at the emergency department with continuous vomiting.

She had started her oncology treatment with pembrolizumab (checkpoint inhibitor) and axitinib (tyrosine

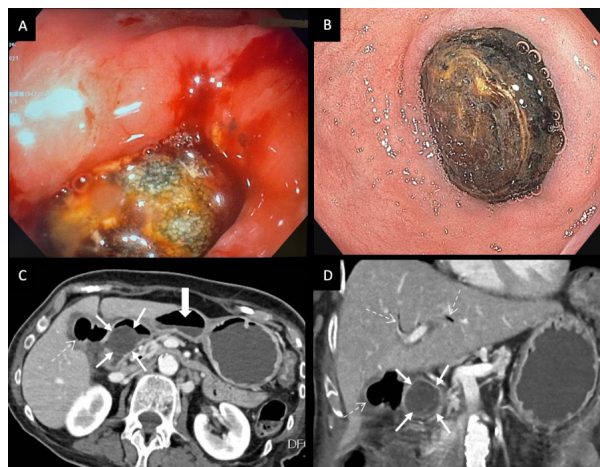


Figure 1. — A + B. Upper endoscopy revealing a large impacted gallstone in the duodenal bulb. C + D. CT scan imaging showing a bilioenteric fistula with air in the gallbladder and pneumobilia (dotted arrow), a large gallstone of 25mm (arrows) impacting the bulb and the stomach filled with fluid and air-fluid levels (large arrow).

kinase inhibitor) 3 months before, which she tolerated well.

One month prior to admission, there had been an incidental and asymptomatic finding of ‘chronic calculous cholecystitis’ on CT with a large gallstone in the gallbladder fundus, as well as small intramural collections. A non-urgent cholecystectomy was scheduled.

At admission she had been vomiting for 5 days and on the day of presentation there was an episode of hematemesis. She was hypotensive and dehydrated and there was an elevated CRP of 137 mg/L (normal range < 5mg/L). Gastroscopic evaluation showed no signs of gastrointestinal bleeding, it did however show a large obstructive, impacted bile stone in the duodenal bulb (figure 1A and 1B). An attempt of endoscopically removing the stone with a snare was unsuccessful.

Additional CT imaging confirmed that the large (2.5 cm) gallstone, previously present in the gallbladder

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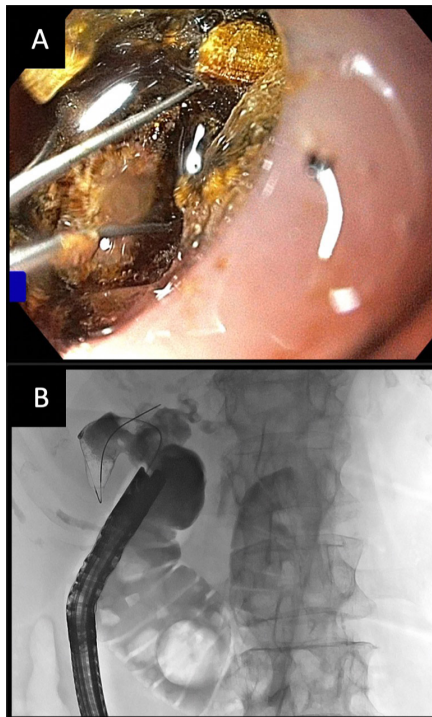


Figure 2. — A. Endoscopic removal of stone fragments with a tripod snare. B. Fluoroscopic image of cannulated gallbladder with no residual stones in the gallbladder lumen.

fundus, had now migrated to the proximal duodenum (figure 1C and 1D). It clearly showed the presence of the bilioenteric fistula between the inflamed gallbladder (with air in the gallbladder lumen and pneumobilia) and the duodenal bulb.

Broad spectrum antibiotics were started and she was transferred to a tertiary center for endoscopic lithotripsy.

Electrohydraulic lithotripsy (EHL) was performed, with the need of 3 EHL probes for full fragmentation of the large gallstone, followed by sequential extraction of the stone fragments with the tripod forceps (figure 2A). After switching to the duodenoscope, the gallbladder lumen could be identified and cannulated with a diagnostic catheter (figure 2B). Considering there was a small residual gallbladder lumen and no residual stones, there was no need for additional therapy.

The procedure was uncomplicated, and oral intake could gradually be increased during the next few days. Given her underlying oncological history, cholecystectomy was not planned.

Discussion

Bouveret syndrome typically presents with symptoms of gastric outlet obstruction. In a review of 128 cases they found 86% to experience nausea and vomiting, 71% had epigastric pain and 27% of patients showed abdominal distention. Other features included hematemesis in 15% and melena in 6% (4).

Diagnosis relies on clinical presentation in combination with imaging and/or endoscopy.

Plain abdominal X-ray is usually non-specific, abdominal ultrasound can show pneumobilia and ectopic location of the gallstone but has various limitations. Computed tomography (CT) scan imaging is most accurate for diagnosing Bouveret syndrome, with a 100% specificity and 93% sensitivity (1). The combination of small bowel obstruction, pneumobilia and the presence of an ectopic gallstone is called the Rigler's triad, which is specific for gallstone ileus (1, 3) and was also present in our case (Figure 1C and 1D).

Upper endoscopy and surgical exploration can have both diagnostic as well as therapeutic purpose. In our case upper endoscopy was performed before any other imaging tool, because of suspicion of upper gastrointestinal bleed. Possible endoscopic findings include a dilated stomach, visualization of the impacted gallstone and/or the cholecystoduodenal fistula.

Surgical exploration is needed for the final diagnosis in 20-40% (3).

There is no consensus or standardized guideline on management, which can be either endoscopic or surgical. Although only successful in less than a third of cases of Bouveret's (3), attemptive endoscopic therapy should still be the primary course of action since it is less invasive than surgery in this population (usually elderly patients with multiple comorbidities).

Endoscopic therapy

With evolving endoscopy techniques, more and more noninvasive methods are becoming available but also require a high degree of expertise. Current strategies for endoscopic treatment of Bouveret syndrome include removal with nets or baskets and various kinds of lithotripsy (mechanical, electrohydraulic, laser and extracorporeal shockwave).

Unsurprisingly, the success rate of removal of an impacted gallstone with a net or basket depends largely on the diameter of the stone. Its use will therefore be limited to smaller stones (generally smaller than 2.5cm), whereas the removal of larger stones will require fragmentation through some form of lithotripsy (2), as seen in our case.

Lithotripsy, or fragmentation, will be necessary for endoscopic removal of larger gallstones, and should always be followed by extraction of the different stone fragments as to reduce the risk of a more distal gallstone ileus caused by migration of any of the fragments (2,3).

Mechanical fragmentation can be carried out using a forceps, basket or snare. If unsuccessful, more specialized lithotripsy modalities are required.

Electrohydraulic lithotripsy (EHL) is a technique initially developed in the industry for fragmenting rocks (5). Its principle is based on creating high-voltage electric sparks at the tip of the EHL fiber, causing spherical shock waves through a surrounding aqueous solution, thereby fragmenting the stone. It is already commonly used for fragmentation of difficult pancreatic and bile duct stones, and can also be successfully applied to treating Bouveret

syndrome, as illustrated in our case. The technique does carry a potential risk of bleeding and perforation caused by damage of the shock waves to the surrounding normal tissue. This risk can be reduced with sufficient water immersion and by avoiding close contact to the intestinal wall (2,6).

Laser lithotripsy is more expensive than EHL, but has a few advantages. First, it can more precisely target the energy onto the stone, therefore causing less injury to the surrounding tissue (2). The energy of laser lithotripsy is not attenuated by intestinal gas, therefore excluding the need for continuous water immersion (7). Some of the lithotripters are even able to differentiate stone from tissue and interrupting signals that may cause tissue damage (2).

Extracorporeal shockwave lithotripsy (ESWL), initially developed for disintegration of renal calculi, can also be a successful treatment for Bouveret syndrome. Shock waves generate compressive stress on the outer surface of the stones, hereby resulting in gradual fragmentation (8). This technique is particularly useful when endoscopic access to the stone is difficult, which is necessary for the other forms of endoscopic fragmentation. ESWL should be followed by endoscopic extraction of the stone fragments.

Surgical therapy

Surgery will be required when endoscopic therapy fails or is unavailable. Surgical treatment options include open procedures (gastrotomy, pylorotomy or enterotomy) as well as laparoscopic enterolithotomy. Laparoscopic procedures are preferred because of the lower morbidity and mortality (2). During surgery, the entire intestine should be carefully inspected for other ectopic gallstones (1,3).

There is still debate on the need for cholecystectomy and fistula repair in combination or following treatment of Bouveret syndrome, for prevention of future biliary complications as well as the occurrence of gallbladder

carcinoma (1). This decision should be made based on the patient's age, comorbidities and life expectancy.

Conclusion

Bouveret syndrome is a rare form of gallstone ileus, presenting with symptoms of gastric outlet obstruction. Diagnosis is based on CT scan imaging (Rigler's triad) and/or upper endoscopy, visualizing a bilioenteric fistula with an impacted gallstone in the pylorus or duodenum. Endoscopic therapy is preferred over surgical intervention, considering that it usually occurs in an older and more frail population. Endoscopic strategy usually involves some form of lithotripsy, followed by extraction of the stone fragments.

Conflict of interest

The authors of this paper report no conflicts of interest.

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