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Bouveret Syndrome: Large Gallstone Causing Duodenal Obstruction

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Abstract **Case Report**

Introduction: Bouveret syndrome consists of a gastric outlet obstruction due to the impaction of a gallstone in the duodenal bulb after it migrates through a cholecystoduodenal fistula. This syndrome represents only 1% to 3% of all cases of gallstone ileus, being more frequent in women and in the elderly. **C*ase report:*** 86-year-old patient with diffuse colicky abdominal pain, diarrhea, and yellowish vomiting. The imaging tests revealed significant duodenal dilation with the presence of a cholecystoduodenal fistula and a large rounded impacted gallstone. The patient underwent emergency surgery through an enterotomy with removal of the gallstone and primary closure. ***Discussion:*** Bouveret's syndrome is a rare gallstone ileus condition that implies significant morbidity and mortality and often occurs in the elderly with significant comorbidities. Individual diagnosis and treatment strategies are required for optimal management and results, being endoscopic treatment or open surgery the two most suitable available options. ***Conclusions*:** Bouveret syndrome is a life-threatening condition with gastric outlet obstruction caused by large gallstones. CT and MRI scans are useful for diagnosis. Although in some cases percutaneous and endoscopic treatments can be successful as first-line treatment, most patients require surgery, however, the decision must be taken individually.

Keywords: Bouveret syndrome, cholecystoduodenal fistula, gallstone ileus, enterotomy.

INTRODUCTION

In 1770, Beaussier first reported gallstone ulcerating in the stomach, causing gastric retention syndrome. Subsequently, in 1896, Leon Bouveret published 2 cases of gastric outlet obstruction due to gallstone impaction in the duodenal bulb occurring through bilioenteric fistulization [1], thus coining his eponymous syndrome.

With only around 300 observations reported in the literature until 2020, Bouveret's syndrome accounts for 1 to 3% of all gallstone related obstruction in the gastrointestinal tract (gallstone ileus), which by itself represents less than 0.1% of cases of small intestinal obstruction in the United States [4]. It is approximately twofold more common in females, with a median age of 74 years at presentation [2].

The pathophysiology of gallstone ileus resembles that of Mirizzi syndrome [8]. Gallstones that are too large to pass through the cystic and common bile ducts (CBD) can result in chronic irritation and inflammation of the gallbladder wall. Over time, this results in gallstone migration through a fistulous tract, creating a cholecysto-duodenal, choledocho-duodenal, or cholecysto-gastric fistula with subsequent obstruction of the upper gastrointestinal tract from gallstone impaction [3].

Mortality rate is estimated between 12 to 30% and reflects the advanced age and fragile state of many of the patients in which it presents. Also, most patients have many comorbidities, making a timely diagnosis and management even more challenging [5, 6].

Common manifestations of Bouveret's syndrome include epigastric pain (71%) and nausea/vomiting (86%); although, other features may include distention (27%), hematemesis (15%), and melena (6%). Physical exam can be unremarkable or can demonstrate abdominal tenderness (44%) and/or signs of dehydration (31%) [7].

CASE REPORT

An 86-year-old woman presented to the emergency department with intestinal occlusion. The patient described hepatic colic for several years, which had intensified recently, associated with fever. Laboratory test results showed inflammatory syndrome, cytolysis, and anicteric cholestasis. A cholangiorresonance was performed, showing a large biliary stone and a cholecysto-duodenal fistula (extending from the gallbladder to the adjacent proximal duodenum). A diagnosis of Bouveret's syndrome was established. Antibiotic therapy (ciprofloxacin + metronidazole) was started, and surgical treatment was performed: a duodenotomy in the lateral anti-mesenteric border of the 2nd part of the duodenum. The defect at the enterotomy site was then sutured by primary closure.

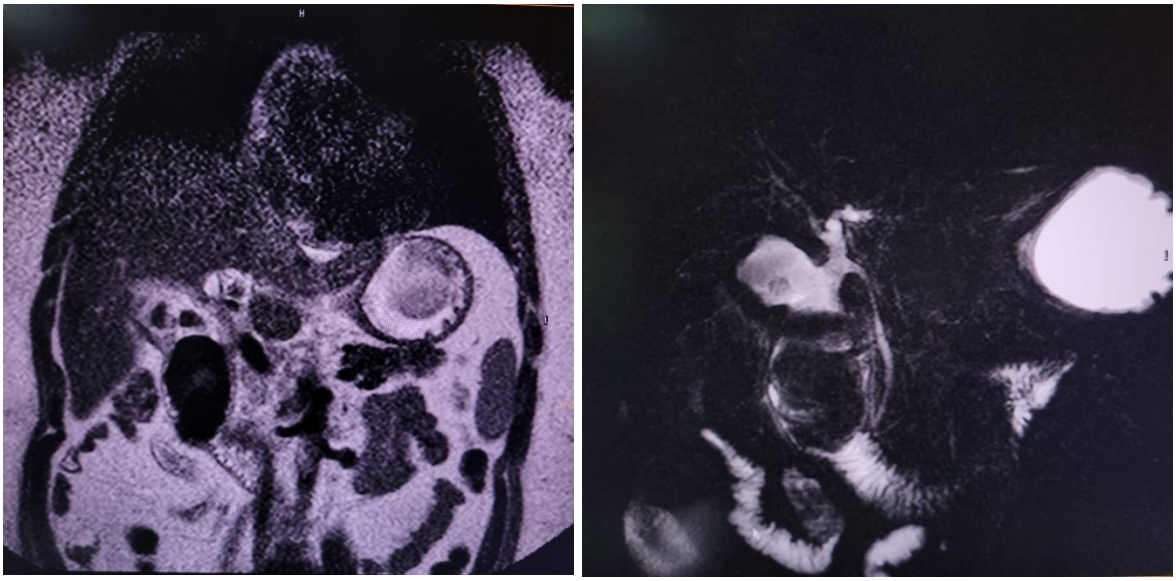

Figure 1: Cholangiorresonance showing large biliary stone and a cholecysto-duodenal fistula

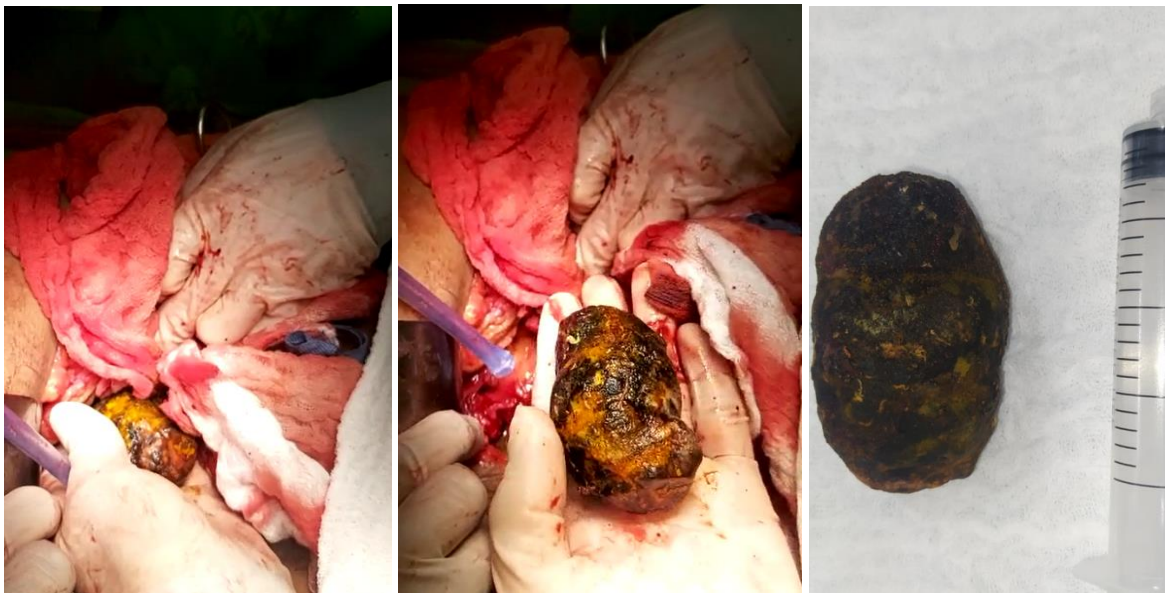

Figure 2: Surgical gallstone extraction and final specimen

DISCUSSION

Luminal complications of errant gallstones occur in less than 1% of cholelithiasis and classically present as gallstone ileus. If obstruction occurs, it most frequently does given the direction of peristalsis and the narrowed caliber of the distal intestine in the terminal ileum [9].

Several endoscopic, laparoscopic, and open surgical techniques have been described in the literature for management of Bouveret's syndrome. However, given rarity of the condition, variable clinical

presentation and complexity of management, there are no specific set of guidelines or protocol to help with diagnosis and management [3]; for this reason, treatment decisions should be made on a case-by-case basis, with an approach for management that should consider candidacy for surgery, medical comorbidities, life expectancy, presence of inflammation and risk of recurrence [14].

Historically, surgery has been the mainstay of treatment for Bouveret's syndrome, with 91% of patients eventually requiring surgery at some point. Success rate has been reported as high as 84% in some case series [2, 7]; and 42% of patients who have required surgery have failed at least one endoscopic trial for removal of stones [2, 8]. Nevertheless, surgical treatment carries an increased risk of morbidity and mortality [1]. Surgery should be considered in hemodynamically unstable patients, patients who fail endoscopic treatment trials, and patients with concurrent distal gallstone ileus. Patients who have undergone endoscopic stone treatment and have stone fragments of more than 1.5 cm, have higher risk of future gallstone impaction and obstruction, so an early surgical management is recommended [10].

Cholecystectomy and fistula repair are recommended to reduce the risk of future recurrence, gallstone pancreatitis, gallstone ileus and gallbladder cancer [4], and they can be performed in one- or in two-stages. Fistula repair options include primary closure, omental patch, jejuonoduodenostomy, and in rare instances, pancreaticoduodenectomy (Whipple) [6, 11].

Abdominal imaging, especially CT imaging is helpful in diagnosis [12]. Although many endoscopic and surgical treatment modalities have been described in the literature, the optimal management of patients with this syndrome can be challenging, given the lack of specific clinical guidelines. Rigler's triad (small bowel obstruction, pneumobilia, and an ectopic gallstone) is found on CT in roughly 78% of cases of gallstone ileus [13].

MRI is also a very useful tool for diagnosing gallstones and associated pathological processes. There is a paucity of literature surrounding MRI/MRCP findings of Bouveret's syndrome with a limited number of case reports and only a single study where the cholecystoduodenal fistula's primary diagnosis was made based on MRI/MRCP [15]. MRI may be especially valuable in demonstrating isoattenuating stones on CT and can also show the extent of the gallbladder's inflammatory process, with the advantage of avoiding associated complications of an invasive endoscopic cholangiography performed for diagnostic purposes [16].

CONCLUSIONS

Bouveret syndrome is a life-threatening condition with gastric outlet obstruction caused by large gallstones. In most cases, a CT scan is required for diagnosis, however, cholangiorresonance is also useful. Although in some cases percutaneous and endoscopic treatments can be successful as a first-line treatment, most patients require surgery to accomplish definitive resolution; anyways, the decision must be taken individually according to the patient's clinical status and resource availability.

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