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Gastric Outlet Obstruction by a Large Gallstone (Bouveret's Syndrome)

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Abstract

Bouveret's syndrome is an uncommon cause of gastric outlet obstruction due to intraluminal duodenal obstruction by a large gallstone, which has migrated via a cholecystoduodenal fistula. The presenting symptoms are not specific in the majority of cases, but most of the patients have known biliary lithiasis. Usually Bouveret's syndrome carries a poor prognosis. Consideration of Bouveret's syndrome in patients presenting with gastric outlet obstruction is important because of the rarity of this entity and the difference in the required surgical approach from that applied for other more common causes of obstruction. We present the case of a 55-year-old male with Bouveret's syndrome, presenting as the first manifestation of gallstone disease, and successfully treated by open surgery, with a comprehensive literature review.

Key words: *Gastric outlet obstruction; gallstone ileus; Bouveret's syndrome; cholecystoduodenal fistula*

Introduction

Bouveret's syndrome is an uncommon cause of gastric outlet obstruction due to a large gallstone, which has migrated through a bilio-enteric fistula. Although only a few sporadic cases had been reported since 1896, when Leon Bouveret first described the syndrome, almost 300 new cases have been reported during the last decade [1-4]. The condition is more common in elderly women with a history of gallstone disease and is associated with high rates of morbidity (60%) and mortality (12-30%) [3,5-9]. An elevated degree of suspicion for Bouveret's syndrome may ensure its early diagnosis, and timely treatment may improve its poor prognosis [10].

We present a case of Bouveret's syndrome that was managed successfully, with a comprehensive review of the literature regarding this entity.

Case Report

A 55-year-old male with no significant past medical or surgical history presented with mild recurrent pain in the upper abdomen for two months, associated with multiple episodes of nausea and non-bilious vomiting.

On admission, the patient had normal vital signs and physical examination showed a non-tender, distended abdomen. Digital rectal examination revealed no abnormalities. Laboratory tests were within the normal range. Abdominal X-ray in the erect position revealed a distended stomach with the appearance of gastroparesis (Figure 1). Abdominal computed tomography (CT) scan depicted a large intraluminal mass in the duodenal bulb, concretions in the gallbladder and a dilated fluid-filled

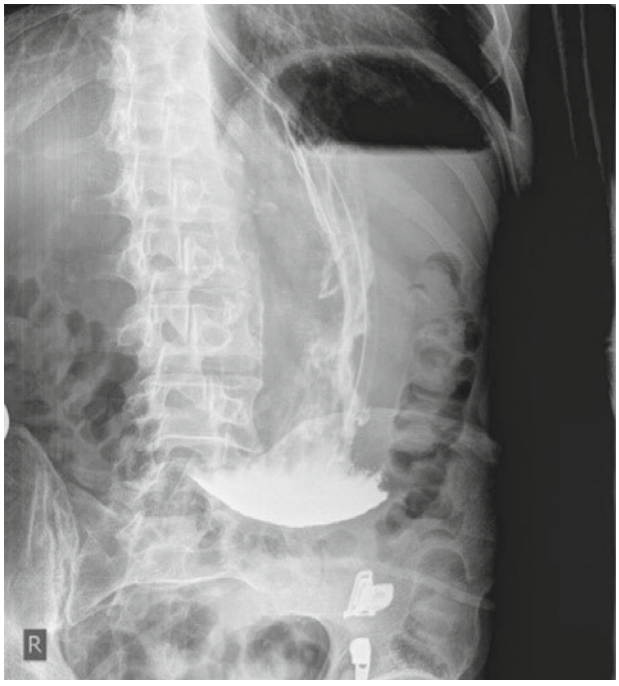

Figure 1. Bouveret's syndrome in a 55-year-old man: Plain abdominal X-ray showing a distended, paretic-like stomach.

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stomach (Figure 2). The patient was admitted with the diagnosis of gastric outlet obstruction. A nasogastric tube was placed to decompress the stomach and the initial treatment included administration of IV fluids with electrolytes and antibiotics.

Upper gastrointestinal (GI) endoscopy revealed gastric outlet obstruction by an impacted gallstone in the duodenal bulb, confirming the diagnosis of Bouveret's syndrome. Attempts to remove the gallstone endoscopically were unsuccessful.

At surgery, access to the abdominal cavity was gained through a midline laparotomy. Intraoperatively a contracted gallbladder was identified, with a fistulous tract connecting it with the first part of the duodenum. Gastrotomy was performed and a 9x4cm gallstone was found impacted in the pylorus (Figure 3). While attempting to remove the gallstone through the gastrostomy, a small perforation of the duodenal bulb occurred. A Petzer tube was inserted through the perforation, which was then closed, while the tip of the nasogastric tube was placed just above it. Subsequently, cholecystectomy was performed and the subhepatic space was drained. The postoperative course was uneventful. Contrast radioscopic examination using Gastrografin® on the 12th postoperative day was negative for duodenal leak (Figure 4). The patient was discharged on the 14th day, since when he returns for regular follow-up.

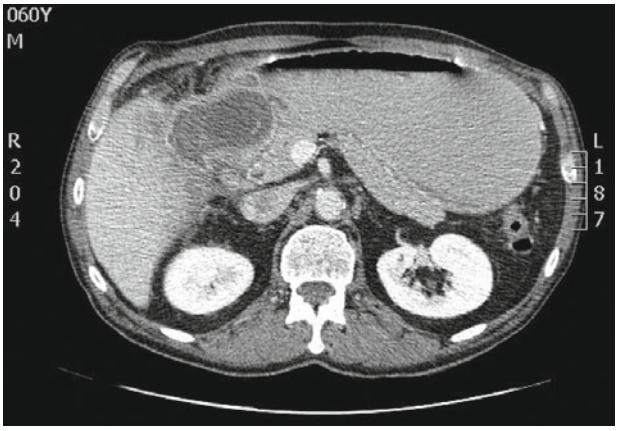

Figure 2. Bouveret's syndrome in a 55-year-old man: Abdominal computed tomography (CT) scan showing a large intraluminal mass in the duodenal bulb and a distended, fluid-filled stomach.

Discussion

Gallstone disease affects 10-15% of the adult population in the USA and Western Europe [6,10]. Less than 20-30% of people with gallstones will develop symptoms [6], and even fewer (1-4%) will develop complications of gallstone disease, which include acute cholecystitis, obstructive jaundice, acute pancreatitis, acute cholangitis and gangrenous gallbladder. Less frequent complications include the Mirizzi syndrome, cholecystocholedochal fistula and small bowel obstruction, referred to as gallstone ileus.

Gallstone ileus is responsible for only 1-4% of all cases of intestinal obstruction and is encountered in 0.3-0.4% of cases of cholelithiasis [7,11,12]. The etiopathogenesis of this condition includes mechanical factors (cystic or common bile duct obstruction, erosion of the gallbladder wall) and septic factors (infection of the gallbladder or digestive

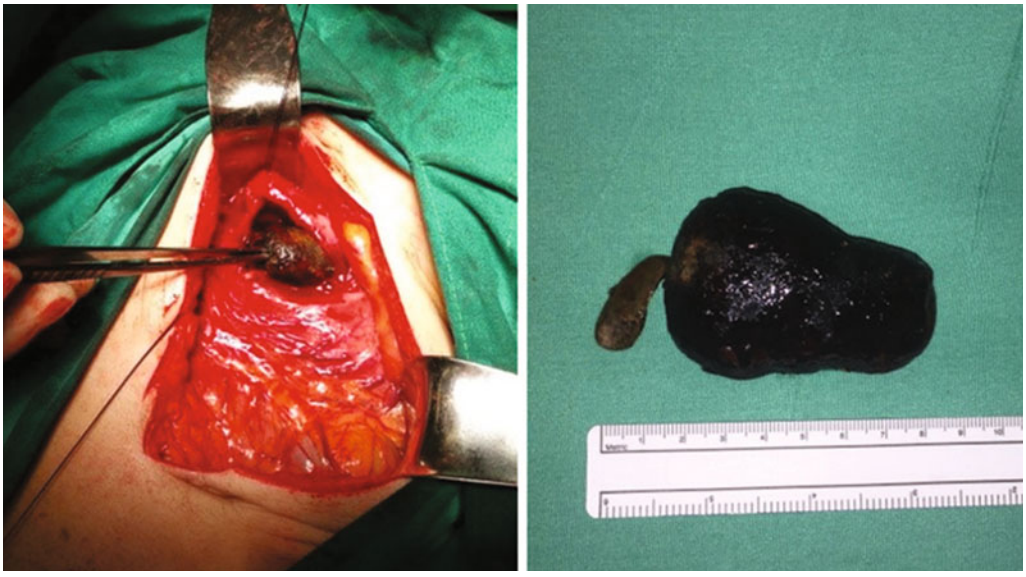

Figure 3. Bouveret's syndrome in a 55-year-old man: Intraoperative pictures showing the obstructing gallstone projecting through the gastrotomy (left) and the removed gallstone (right).

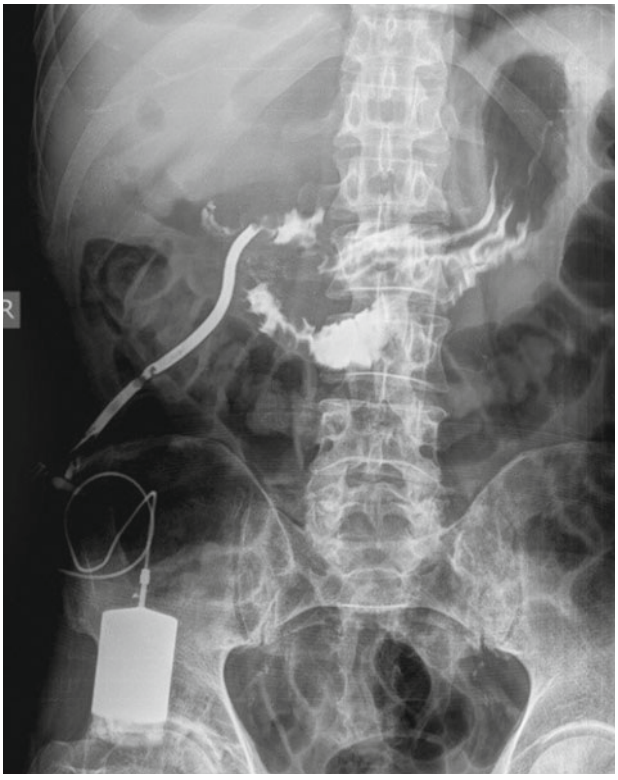

Figure 4. Contrast radioscopic examination on the 12th postoperative day, negative for duodenal leak.

tract). Gallstone ileus is significantly more common in elderly females and patients with diabetes mellitus (DM) and atherosclerosis [13]. The gallstones migrate through a bilio-enteric fistula into the GI tract and become impacted in the bowel, causing bowel obstruction. The biliary-digestive fistula is most commonly cholecystoduodenal (60%) [14]; less common variants are cholecystocolic (17%), cholecystogastric (5%) and choledochoduodenal (5%). The most common site of obstruction is the terminal ileum (60-70%), followed by the proximal ileum (25%), distal ileum (10%), jejunum (9%), colon (4%), rectum (2%) and duodenum (1-3%) [1,5,12-14]. As a general rule, the larger the gallstone, the more proximal the obstruction [7,11]. Sometimes the gallstone may enlarge on its passage through the fistula and the intestine, due to accumulation of salts and fecal material on its surface [3].

Bouveret's syndrome is an uncommon cause of gastric outlet obstruction, due to intraluminal duodenal obstruction by a large gallstone, that has migrated through a bilio-enteric fistula. The pathophysiology of all cholecystoenteric fistulae is similar; chronic luminal pressure from a large gallstone results in localized necrosis and perforation through the wall of adjacent bowel [6,13].

The clinical presentation of Bouveret's syndrome is nonspecific. Common symptoms are nausea, vomiting, abdominal pain or discomfort, recent weight loss, anorexia, fever, early satiety, hematemesis and melena [3,6,15,16]. More than 60% of the reported cases have an associated previous clinical history, and the majority of patients have known biliary lithiasis, without excluding the possibility of Bouveret's syndrome being the first manifestation of gallstone disease, as in our case. Common signs of Bouveret's syndrome are abdominal tenderness, signs of dehydration, abdominal distention, obstructive jaundice and pyrexia. Laboratory studies may demonstrate leukocytosis and electrolyte and acid-base disorders. Less commonly, abnormal liver chemistry indices, including raised total and direct bilirubin, alanine aminotransferase (ALT), γ-glutamyl transferase (γGT) and serum amylase are observed. Preoperative diagnosis is based on imaging studies. Rigler's triad describes the radiographic findings most classically associated with gallstone ileus and consists of small bowel obstruction, pneumobilia and an aberrant stone in the GI tract seen on imaging [6,12,17,18]. Plain abdominal X-ray in Bouveret's syndrome is diagnostic in about 30-50% of cases [12,19] and may demonstrate gas in the biliary tract [12,13] and gastric dilatation [12]. Ultrasonography (U/S) may reveal gallstones in 75% of cases of Bouveret's syndrome (23% of which are in the duodenum), pneumobilia in 45% of cases [6], gastric dilatation and the pathognomonic "double arch sign" [20].

Abdominal CT scan, which is currently the preferred imaging modality, may reveal pneumobilia, migrated gallstones, cholecystoduodenal fistula and a contracted gallbladder [6,18,19,21,22]. Rigler's triad is present on abdominal CT scan in 78% of cases [12,13]. Oral and IV contrast improves the diagnostic sensitivity of CT scan, as it surrounds the gallstone [18,23,24]. Magnetic resonance cholangiopancreatography (MRCP) is a useful diagnostic tool for cholecystoduodenal fistula [17,18], as it does not require the use of oral contrast, but it remains secondary to CT scan. Bouveret's syndrome was first diagnosed by endoscopy of the upper GI tract in 1976 [25] and gastroscopy continues to be an important adjunct to diagnosis and can be therapeutic [3,18,21]. Common endoscopic findings are: impacted stone causing obstruction (69%), obstruction without visualized stone or fistula (31%), stone partially visualized through duodenal wall (<1%), inflammation, edema or ulcer at site of impaction and retained food or fluid in the stomach [21].

Bouveret's syndrome must be distinguished from other causes of gastric outlet obstruction, such as gastric cancer and peptic stenosis [19]. Duodenal compression by a massively enlarged gallbladder containing multiple calculi may also cause gastric outlet obstruction (pseudo-Bouveret's syndrome) [26]. A gallstone in the intramural submucosal portion of the duodenum has also been reported as a pos-

sible cause of obstruction [27]. Rarely, Bouveret's syndrome results in complications such as duodenal perforation, hematemesis (Mallory-Weiss tear), distal esophageal rupture (Boerhaave's syndrome) and gastric bezoar [21,28].

The optimal treatment of patients with Bouveret's syndrome is still a subject of debate in the world literature. The primary goal is to resolve the obstruction by removing the impacted stone. This can be achieved endoscopically or surgically (open or laparoscopic approach). Endoscopic removal of the impacted stone should be the initial approach, if feasible [22]. Endoscopic management, when possible, has low morbidity and negligible mortality. The presence of an experienced endoscopist is a prerequisite, but even then, endoscopic removal of the stone can be achieved in less than 10% of cases [2,11,19,29,30]. When the calculus is very large, endoscopy often fails. as in our case, where the size and shape of the gallstone (9x4cm, pear-shaped) obstructing the gastric outlet made endoscopic management impossible. While the majority of patients tolerate endoscopic treatment, pulseless electrical activity may occur during mechanical retrieval, due to the gallstone becoming lodged in the esophagus [5]. Another possibility that must be taken into consideration is that fragments of the initial gallstone may migrate to the distal part of the intestine, leading to a new obstruction. Indications for open surgery are: stone size greater than 2.5cm, residual stones in the gallbladder, multiple stones in the intestinal lumen, sepsis, perforation, stricture and failure of the endoscopic approach [3,9,12]. Open surgery entails removal of the obstructing stone via enterotomy or gastrotomy, followed by cholecystectomy and fistula repair. Many authors suggest a more conservative surgical approach consisting of removal of the obstructing gallstone, with or without cholecystectomy and without fistula repair, especially in elderly, high risk patients [6,14,27,28,31]. This approach is supported by studies showing that cholecystectomy and surgical management of the fistula carry a higher mortality and complication rate than when the gallbladder and fistula are left *in situ*. There is documentation of spontaneous closure of the fistula when the cystic duct is left open following obstructive gallstone removal [7,22,31]. Fistula removal under difficult dissection conditions may call for an incomplete cholecystectomy with a reduced rate of biliary duct injury [13]. In our case the indication for open surgery was the size of the stone and resultant failure of the endoscopic approach. During the procedure, at the time of the extraction of the impacted gallstone, the fistula tore apart and a small rupture of the duodenal bulb occurred, rendering the decision for one-stage operative management imperative. Cholecystectomy was not easy because of the adhesions of the gallbladder, but all the anatomical structures were safely identified.

Some authors favor a one-stage operation with simultaneous enterolithotomy, cholecystectomy and fistula repair [18], which is associated with less biliary complications, but with a higher mortality rate than the two-stage operation (20-30% vs 12%) [14]. If the fistula opening is small, primary repair can be attempted. Otherwise gastric or duodenal resection and gastroenteric anastomosis should be performed [7,32,33]. Many surgical centers now attempt laparoscopic treatment of Bouveret's syndrome [3,6,9,34].

Despite the advances in endoscopic technique, which is commonly the initial approach, the prognosis of Bouveret's syndrome is poor and the mortality rate is 15-18%. This is justified by the low percentage of successful endoscopic treatment (less than 10%) and the co-existence of bilioenteric fistula, which poses significant technical challenges for the surgeon. The morbidity associated with endoscopic treatment itself is not always negligible, given the possibility of distal bowel obstruction by stone fragments and duodenal injury caused by vigorous endoscopic manipulations [6,8, 11,13]. Finally, cases with late diagnosis, in patients of older age, and with obesity and DM, carry a worse prognosis.

In conclusion, Bouveret's syndrome is a rare complication of a fairly common entity, i.e., gallstone disease. It carries a poor prognosis, making its overall therapeutic approach a true challenge for every clinician. It is a condition that should be considered as part of the differential diagnosis of gastric outlet obstruction. This presentation of one more case of Bouveret's syndrome is intended to enrich the scarce literature, and to demonstrate the feasibility of a one-stage operation, consisting of simultaneous removal of the gallstone obstructing the gastric outlet, fistula repair and cholecystectomy.

Conflict of Interest: *The authors declare that they have no conflict of interest.*

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