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Case Report

Bouveret Syndrome Presented with Upper Gastrointestinal Bleeding and Complicated by Distal Gallstone Ileus

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Abstract

Bouveret syndrome is a rare variant of gallstone ileus. It is defined as a gastric outlet obstruction caused by impaction of a gallstone to the stomach or the duodenum through a cholopeptic fistula. It was reported in elderly patients with multiple comorbidities. Due to lack of specific signs and difficulties in visualization of the stone, the diagnosis is usually delayed. We present an 81-year old man with Bouveret syndrome presented with severe hematemesis and complicated by distal gallstone ileus, who was managed successfully with gastrotomy, enterectomy with stone removal and gastrointestinal anastomosis.

Keywords: *Bouveret Syndrome; Upper Gastrointestinal Bleeding; Distal Gallstone Ileus*

Introduction

Bouveret syndrome is a rare complication of cholelithiasis with high morbidity and mortality rates [1]. The term is used to describe the gastric outlet obstruction caused by a gallstone impacted in prepylorus/pylorus or the duodenum and it is thought to account for 1 - 3% of gallstones ileus cases [1]. The formation of a cholopeptic fistula via which the gallstones pass to the gastrointestinal (GI) lumen is the prerequisite condition [1]. The epidemiological characteristics of the most commonly affected population are the advanced age, female gender, history of chronic symptomatic cholelithiasis and significant comorbidities [1].

We herein present a case of Bouveret syndrome admitted with upper gastrointestinal bleeding who developed distal gallstone ileus during hospitalization.

Case Report

An 81-year-old male presented at the emergency department with 2 episodes of hematemesis. His past medical history included coronary artery disease, heart failure and arterial hypertension. His medication included clopidogrel, atorvastatin, carvedilol, furosemide and pantoprazole. At admission, his vital signs were normal and the abdominal examination was unremarkable. Laboratory investigations revealed hemoglobin levels of 10 gr/dl, urea of 77 mg/dl (normal range 15 - 40 mg/dl), a white blood cell count of 11.94 x 10^9 cells/L and C-reactive protein of 19 mg/dl (normal < 5 mg/dl). The remaining laboratory investigations were normal. The plain X-ray of chest and abdomen were normal at that time. The patient was initially managed with intravenous fluids, nasogastric tube and withdrawal of clopidogrel. An upper gastrointestinal (GI) endoscopy at admission revealed "a mass" obstructing the pylorus. Two episodes of melena with concomitant hemodynamic instability ensued two days later and he was transfused. As there was a strong possibility of underlying gastric malignancy, a second upper GI endoscopy was arranged for biopsies to be obtained. The second endoscopy revealed a large gallstone impacted in a fistulous stoma at the pylorus (Figure 1A), causing partial obstruction of the gastric outlet. Multiple gastric erosions were also seen. Gallstones of smaller diameter (< 1 cm) were visible at the antrum of the stomach (Figure 1B). Attempts to push, remove or bypass

the stone were unsuccessful. The surgeon then suggested operative management. On the third day following admission, his abdomen rendered distended and diffusely tender and the plain abdominal X-ray was compatible with small bowel obstruction. The computed tomography (CT) of the abdomen demonstrated air in the gallbladder (pneumobilia) (Figure 2A) and multiple calculi within the duodenum and jejunum (Figure 2B-2D). Patient underwent exploratory laparotomy. An enterectomy was performed and four firmly impacted gallstones, at 30 cm beyond the Treitz ligament, were eventually removed from the jejunum (Figure 3A). A jejuno-jejunal anastomosis hand-sewn in two layers was performed. In addition, the impacted gallstone in the stomach was removed by a gastrotomy (Figure 3B). The procedure was completed by gastro-jejunal anastomosis to prevent gastric outlet obstruction due to edema in the region of the fistula. The diagnosis of Bouveret syndrome complicated with upper gastrointestinal bleeding and distal gallstone ileus was set.

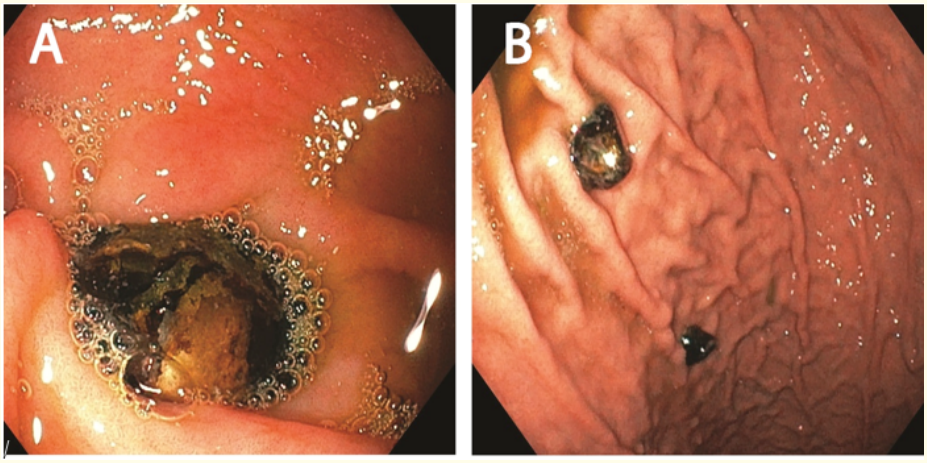

Figure 1: *(A) A large gallstone impacted in a fistulous stoma at the pylorus (B) small gallstones at the antrum of the stomach.*

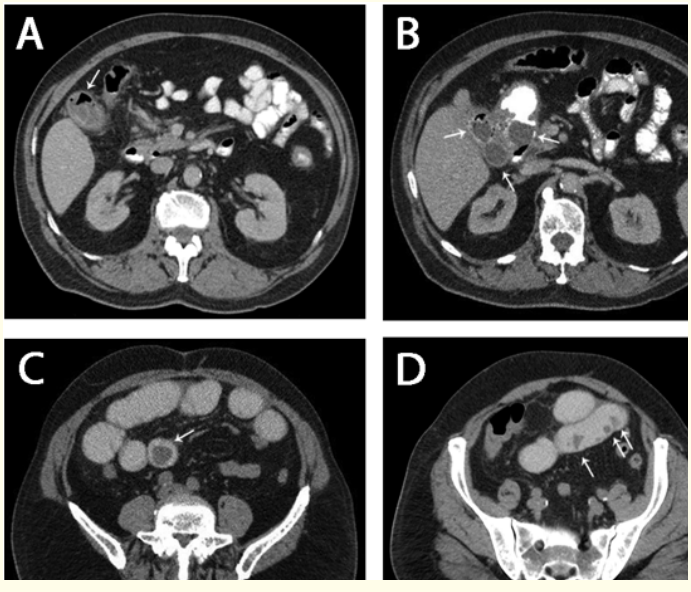

Figure 2: *(A) Air in gallbladder (pneumobilia) (arrow) and (B) Multiple calculi in the duodenum (C, D) multiple calculi within the loops of the jejunum (arrows).*

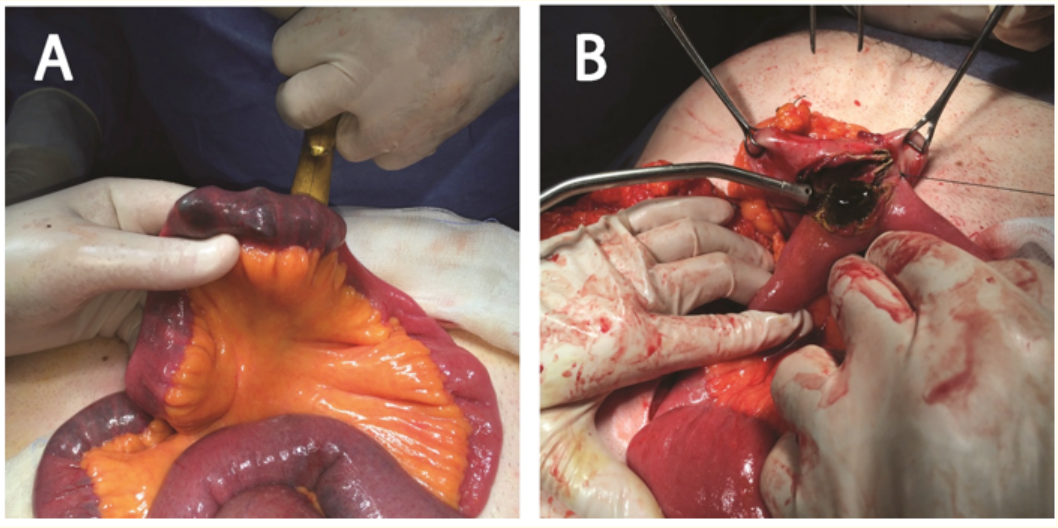

Figure 3: *(A) Jejunum at 30 cm beyond Treitz ligament with four impacted stones. Enterectomy was performed (B) Gastro-jejunal anastomosis. Extraction of a gallstone from duodenum through the gastrotomy.*

The postoperative course was uneventful and the patient was discharged home on postoperative day 10. Five months following the procedure, the patient is still asymptomatic.

Discussion and Conclusion

Gallstone ileus is a rare complication of cholelithiasis with a female to male ratio of 3.5 - 6:1 [1]. It is associated with a cholopeptic fistula which allows the passage of a large gallstone to the digestive tract [1]. In most cases, fistula originates from the gallbladder and seldom from the common bile duct [1]. The diameter of the stone must be at least 2 - 2.5 cm in order to cause obstruction [2]. This rare disorder accounts for 1 - 4% of bowel obstruction cases in the general population but it may increase to 25% in patients older than 65 year-old [3]. Gallstones can be impacted in any part of the GI tract. According to Reisner., *et al*. [4] impaction is most prevalent in the ileum and the ileocecal valve (60.5%) because of their narrow lumen. Less common locations are the jejunum (16.1%) as in present case, the stomach (14.2%), the colon (4.1%), and the duodenum (3.5%) [3].

Bouveret syndrome, comprising 1 - 3% of cases of gallstone ileus, is a gastric outlet obstruction by a stone impacted in the prepylorus/pylorus or the duodenum [5]. According to a review of 128 cases, the clinical picture is characterized by nausea/vomiting (86%), abdominal pain (71%), recent weight loss (14%), anorexia (13%) and melena (6%) [1]. Hematemesis, as the initial manifestation of Bouveret syndrome, accounts for only 15% [1] of clinical manifestations. The absence of the signs and symptoms of gallstone disease as in the present case, does not exclude this entity as in up to 27% of cases, gallstone disease may be asymptomatic [1].

Despite the wide variety of the available diagnostic methods, Bouveret syndrome remains a challenging diagnosis. Rigler's triad consists of pneumobilia, ectopic gallstone and obstruction [6] and is thought to be pathognomonic for Bouveret syndrome. However, it is evident in only 15 - 30% of plain abdominal X-rays [1,7]. Ultrasound reveals the presence of pneumobilia or gallstones in 45% or 75% of cases, respectively [7]. Moreover, gallstone or fistula are visible in upper GI endoscopy in 69% or 13% respectively [1]. In the remaining cases, findings are misleading [8], resulting in a significant delay in diagnosis [1,8,9]. CT scan is the preferred imaging technique illustrating Ringler's triad in 77% of cases [7,10]. However, the diagnosis is confirmed perioperatively in almost 50% of patients [2].

The coexistence of Bouveret syndrome with classical distal gallstone ileus in the present case is outstanding and has rarely been described in the literature [10]. It may have been resulted either as a consequence of the previous passage of small gallstones to distal small bowel or induced iatrogenically by multiple attempts to remove the large gallstone from the pylorus during endoscopy [10].

There is no consensus on the optimal treatment for gallstone ileus. Less invasive, endoscopic methods including mechanical lithotripsy, electro-hydraulic lithotripsy, laser lithotripsy, net extraction or combinations of the above are preferred due to their significantly lower morbidity and mortality. Unfortunately, endoscopic treatment is successful in only 9% [1,11,12]. More than 90% of patients will eventually need surgery. The best surgical approach is still controversial. There are two surgical options. The first two-stage approach includes urgent enterolithotomy and/or gastrotomy alone followed by cholecystectomy and fistula closure at a later date. The second one is a single stage approach combining both aforementioned procedures in the same time [7]. Mortality rate is 12% for the former and 17% for the latter, respectively [7,13]. The former is preserved for elderly patients with significant comorbidities and carries a risk of recurrence of the gallstone ileus and of complications from biliary system. An alternative choice is laparoscopic surgery in selected patients [13]. An individualized approach may be the most appropriate method of treatment.

In conclusion, Bouveret syndrome is still a medical condition with high mortality. Elderly patients with multiple comorbidities are the population at risk. Delay in diagnosis due to both nonspecific symptoms and failure of imaging and endoscopy methods to recognize it, is common. Bouveret syndrome presented with hematemesis and complicated by distal gallstone ileus is a diagnostic and surgical challenge but it may be successfully managed by experienced surgeons.

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