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Bouveret Syndrome – A Diagnostic Dilemma

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ABSTRACT

Bouveret syndrome referred to a rare form of gastric outlet obstruction caused by a large gallstone impaction into the duodenum or the gastric pylorus. Our case represents diagnostic dilemma of Bouveret syndrome with uncertainty with diagnosis, failed endoscopic approach, non-diagnostic percutaneous core biopsy, with subsequent major exploratory operation and postoperative complications and favorable outcome.

KEYWORDS: Gallstone ileus; Gastric outlet obstruction; Bilioenteric fistula; Bouveretsyndrome; Cholelithiasis.

INTRODUCTION

Bouveret syndrome is rare form of gallstone ileus when a gallstone is impacted in the duodenum or the distal stomach via bilioenteric fistula and causes gastric outlet obstruction. It is more common in the elderly and in females with a reported median age of 74 years [1]. The formation of a fistula is favored by the long history of cholelithiasis, repeated attacks of acute cholecystitis, a large stone and advanced age [3].

Gallstones obstructing the duodenum are usually more than 2.5 cm in size. Prevalence of bilioenteric fistulas is estimated to be between 0.3 and 5% amongst patients with cholelithiasis and it is seven times more frequent in patients aged over 70 [2].

An early diagnosis of Bouveret's syndrome might be challenging due to variable and non-specific presentation. A triad of epigastric pain, nausea and vomiting are the most commonly described symptoms. Abdominal distention and dehydration are the other common, non-specific symptoms. Hematemesis is rare and it is secondary to duodenal and celiac artery erosion [1]. Differential diagnosis includes conditions cause gastric outlet obstruction such as stomach cancer, peptic stenosis, gastric volvulus and bezoars.

DIAGNOSIS

The most sensitive diagnostic imaging modality is CT scan. The elements of the Rigler's triad seen on the scan are bowel obstruction, pneumobilia and an ectopic gallstone. Approximately 25% of gallstones are missed on CT scan due to radiolucency. Abdominal plain

X ray offers less diagnostic value due to unspecific findings [3]. Ultrasonography might be valuable in some cases, however, is often limited due to anatomical changes such as dilated bowel, contracted or air-filled gallbladder. If patient can tolerate oral intake, a barium meal could also be helpful. In cases with radiolucent stones, MRCP is valuable, as it distinguishes stones and fluid and can visualize the fistula. ¹ In about 70% cases, the impacted stone can be visualized on gastroduodenoscopy and removal of the stone can be attempted same time, however the success rate is minimal [3]. Despite all diagnostic modalities, in 20-40% cases the final diagnosis is established during surgery.

TREATMENT

Treatment options include endoscopy and surgery. Removal of impacted stone with endoscopic technique is usually considered as a first line, however, is challenging, particularly with larger stones, and can be achieved with mechanical, laser lithotripsy and electrohydraulic modalities [1]. In case of failed endoscopic approach, surgery is the mainstream treatment. Laparoscopic techniques should be considered whenever possible to minimize operative trauma, however the reported conversion to open rate is rather high due to complexity.

Our case represents diagnostic dilemma of Bouveret syndrome with uncertainty with diagnosis, difficult anatomy and subsequent major exploratory operation with postoperative complications, and favorable outcome.

CASE REPORT

A 72 years old Mrs. W. with history of diabetes presented with 4 weeks' history of vomiting after each meal and weight loss. No abdominal pain was reported. On examination her vitals were stable, abdomen was soft, nondistended and nontender. Laboratory results showed leukocytosis with neutrophil count $12.4 \times 10^9/L$, mild increase in acute phase inflammatory markers (C reactive protein of 34), unremarkable electrolytes and normal renal function. The patient was referred for an urgent outpatient gastroscopy.

On gastroscopy a large amount of bilious fluid in stomach and an intrinsic severe, non-traversable pyloric stenosis were identified. Subsequent biopsies of the stenotic area shown inflamed gastric mucosa with focal intestinal metaplasia.

An abdominal Computed Tomography (CT) revealed a soft tissue density mural mass 6.7 cm x 5 cm, with dense calcification involving the pyloric antrum, pyloric canal, and duodenal cap with subsequent pyloric obstruction. The possibility of GIST, carcinoma of the pylorus or gallbladder were suspected as well as migrating gallstone. CT scan guided biopsy of the pyloric antrum region was performed with no diagnostic features of GIST or occult carcinoma identified. MRI scan was not performed at the time (Figure 1).



Figure 1: CT scan showing a large soft tissue density mural mass with dense calcification involving the pyloric antrum, pyloric canal, and duodenal cap with subsequent pyloric obstruction.

Considering a wide range of differentials, the patient was consented for laparotomy with resection of distal stomach, first part of the duodenum and cholecystectomy. Intraoperatively, gallbladder was found to be contracted and scarred, with a difficult Calot's triangle dissection. Intraoperative cholangiogram was obtained to reconfirm anatomy prior to cholecystectomy. No bilioenteric fistula was visualized. 50% distal stomach with proximal duodenum were resected en-bloc. A Roux-en-Y reconstruction was performed with gastrojejunostomy and enteroenterostomy with a stapled anastomosis. Patient was transferred in high dependency unit post operatively (Figure 2, 3, 4).



Figure 2: Intraoperative cholangiogram showing contrast in the CBD with flow through the duodenum. No discrete filling defect or bilioenteric fistula visualized.

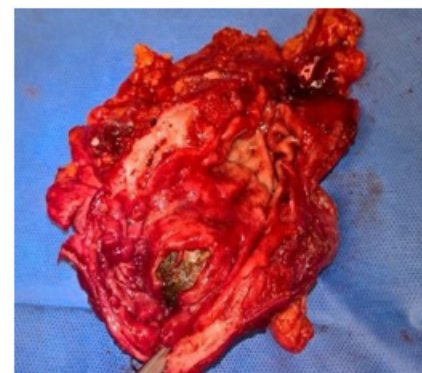


Figure 3: Intraoperative specimen. Distal stomach with proximal duodenum. A gallstone impacted in the duodenum with associated ulceration and inflammatory changes. Histology shown adjacent perigastric fibrosis consistent with healed perforation.



Figure 4: Intraoperative specimen. Distal stomach with proximal duodenum and impacted large gallstone (manually disimpacted in vitro)

Postoperative course was complicated by localized liver collection, secondary to probable duodenal stump leak that required CT guided percutaneous drainage, multiple courses of antibiotics and total parental nutrition. Furthermore, patient suffered from anastomotic bleed requiring blood transfusion. Overall recovery was lengthy and troublesome. Patient was discharged home after 40 days with abdominal drain in situ. The drain was subsequently removed 2 weeks after hospital discharge and the repeat CT confirmed no residual collection.

DISCUSSION

Bouveret syndrome is rare form of gastric outlet obstruction caused by a large gallstone and its diagnosis can be challenging. It is commonly seen in advanced age female patients and symptoms often not specific. In the before CT era Bouveret syndrome was associated with high mortality rate of 30%, partially due low sensitivity diagnostic modalities. Even these days, in some cases it can implicit a diagnostic dilemma. In 20-40% of cases, diagnosis of gallstone ileus is finally established intraoperatively.¹ This case report shows how relatively benign but rare and under diagnosed pathology can lead to a large operation with prolonged, difficult postoperative recovery.

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