Duodenal obstruction by a gallstone (Bouveret's syndrome) managed by endoscopic stone extraction: A case report and review

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F Schweiger, R Shinder. Duodenal obstruction by a gallstone (Bouveret's syndrome) managed by endoscopic stone extraction: A case report and review. *Can J Gastroenterol* 1997;11(6):493-496. Gastric outlet obstruction caused by a large gallstone in the duodenum or pylorus (Bouveret's syndrome) is a rare complication of gallstone disease. The presenting symptoms are often nonspecific and include nausea, vomiting, epigastric pain and a history of gallbladder disease. Although the diagnosis is established only at surgery in many cases, preoperative recognition by imaging techniques and endoscopy is desirable. Surgical treatment aims at removal of the ectopic gallstone, closure of the fistula and cholecystectomy. A case of Bouveret's syndrome is presented where endoscopic extraction of the duodenal gallstone was accomplished providing definitive treatment for this patient.

Key Words: *Bouveret's syndrome, Cholecystoduodenal fistula, Endoscopic stone extraction*

Obstruction duodénale par calcul biliaire (syndrome de Bouveret) traitée par extraction endoscopique : rapport de cas et revue de la littérature

RÉSUMÉ : L'obstruction du bulbe duodénal causée par la présence d'un calcul biliaire dans le duodénum ou le pylore (syndrome de Bouveret) est une complication rare de la lithiase biliaire. Les symptômes typiques sont souvent non spécifiques : nausées, vomissements, douleurs épigastriques et antécédents de maladie vésiculaire. Bien que le diagnostic ne soit confirmé qu'à la chirurgie dans de nombreux cas, la reconnaissance préopératoire au moyen de techniques d'imagerie et d'endoscopie est souhaitable. Le traitement chirurgical vise l'élimination du calcul ectopique et la fermeture de la fistule et englobe la cholécystectomie. Nous présentons ici un cas de syndrome de Bouveret où l'extraction endoscopique du calcul biliaire duodénal a été réalisée, offrant de ce fait un traitement définitif chez le patient.

Bouveret's syndrome is a rare complication of gallstone disease, with fewer than 200 cases described in the literature (1). It is defined by the formation of a cholecystoduodenal or choledochoduodenal fistula with passage of a gallstone into the duodenal bulb or pylorus leading to gastric

outlet obstruction. The clinical diagnosis is frequently missed preoperatively, and until recently, surgery was considered to be the only possible treatment (2).

We describe a patient with Bouveret's syndrome who was diagnosed and successfully treated endoscopically.

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Figure 1) Endoscopic appearance of gallstone in the duodenum

CASE PRESENTATION

An 83-year-old man presented with a two-week history of epigastric and right upper quadrant pains associated with anorexia, nausea and mild weight loss. Eventually he developed recurrent vomiting necessitating admission to hospital. He denied fever, jaundice, hematemesis and melena.

His past medical history included advanced ischemic heart disease, mild renal insufficiency, chronic normocytic normochromic anemia and gout, as well as a 40-year history of ulcerative colitis. Two years earlier, a cecal tubulovillous adenoma was removed by colonoscopy, and inactive universal ulcerative colitis was confirmed with no evidence of dysplasia on multiple random biopsies. Incidental cholelithiasis and an otherwise normal biliary tree were shown by abdominal ultrasound at that time. His medications consisted of enteric coated acetylsalicylic acid, captopril, furosemide, allopurinol and iron tablets. He did not smoke or drink alcohol.

On examination he looked pale and unwell but was stable hemodynamically with no signs of heart failure or jaundice. His temperature was 38.2°C. Mild right upper quadrant tenderness without rebound tenderness could be elicited. There were no masses or organomegaly. Rectal examination suggested prostatic hypertrophy. Initial laboratory studies revealed hemoglobin 73 g/L (normal 133 to 169 g/L), normal white blood cell count, alkaline phosphatase 505 U/L (normal 39 to 117 U/L), alanine aminotransferase 45 U/L (normal 0 to 40 U/L), aspartate aminotransferase 62 U/L (normal 7 to 37 U/L), total bilirubin 10 µmol/L (normal 4 to 18 µmol/L), serum creatinine 219 µmol/L (normal 60 to 120 µmol/L) and normal electrolytes. Evaluation of his anemia showed normal serum B12 and red cell folate levels. The results of iron studies agreed with those for anemia of chronic disease.

An ultrasound showed a slightly thickened gallbladder wall, cholecystolithiasis and air in the intrahepatic bile

Figure 2) Gallstone along greater curvature of gastric body

ducts. An endoscopic retrograde cholangiopancreatography revealed a small amount of fresh blood in the stomach in the absence of any esophageal or gastric lesions. Intubation of the duodenum was prevented by a large gallstone impacted in the first part of the duodenum (Figure 1).

The stone was pulled back into the stomach by using a polypectomy snare (Figure 2). The duodenoscope was then advanced into the duodenum. An oval shaped ulcer was noted in the duodenal apex with a small fistulous opening draining green bile when endoscopic suction was applied (Figure 3). The proximal portion of the descending duodenal limb appeared stenotic but permitted passage of the duodenoscope (Figure 4). The papilla appeared normal, and the pancreatogram was unremarkable. Upon cannulation of the fistula, a cholangiogram was obtained, confirming air in the biliary tree, no evidence of ductal abnormalities and a patent cystic duct. The gallbladder could not be outlined fully due to back-leakage of contrast medium through the large fistulous orifice, as well as some extravasation of hypaque around the gallbladder suggesting a localized rupture of the cystic duct and, less likely, the gallbladder wall (Figure 5). Thereafter, with the use of a forward viewing gastroscope, the stone in the gastric lumen was grasped with a snare once more and removed orally together with the endoscope (Figure 6), encountering moderate resistance at the level of the cricopharynx. The stone was smooth and measured 2.7 cm in maximum diameter (Figure 6).

The patient was transfused and placed on a 10-day course of intravenous antibiotics, as well as oral omeprazole 40 mg/day. His symptoms completely disappeared, and his liver enzymes normalized three weeks later. Five months later he remained asymptomatic and had gained 6.8 kg. A repeat ultrasound showed cholelithiasis and a thickened gallbladder wall without evidence of bile duct dilation or intrabiliary air.

Figure 3) Oval-shaped ulceration surrounded by marked erythema and friability. Fistulous opening (arrowhead)

Figure 4) Stenotic proximal portion of descending duodenum. Cannula points towards cholecystoduodenal fistula (arrow)

DISCUSSION

Biliary-enteric fistulas form in 0.3% to 0.5% of all patients with gallstones, accounting for 1% to 3% of cases of mechanical small intestinal obstruction (3). A fistulous communication occurs between the gallbladder (or rarely the bile duct) and the duodenum in approximately 70% of cases. Fistulas into the colon, jejunum and stomach occur much less frequently (4,5).

Figure 5) Endoscopic retrograde cholangiopancreatography showing air in biliary tree (arrow) and contrast in the subhepatic space (arrow-head)

Figure 6) The recovered gallstone with a smooth green-black surface

Acute pericholecystic inflammation results in the formation of adhesions between the gallbladder and the intestine. Most often, large solitary stones cause a chronic inflammatory process leading to localized ischemia of the gallbladder wall. As intraluminal pressure increases, necrosis of the biliary-digestive barrier ensues resulting in the formation of a fistula (6).

Stones less than 2.5 cm in diameter usually pass spontaneously, whereas larger stones lead to obstructive symptoms because they lodge in the terminal ileum in 73%, and in the proximal ileum or jejunum in 14% of cases (5). Duodenal obstruction is rare and occurs in about 3% of patients with gallstone ileus (3).

As in our patient, the clinical presentation of Bouveret's syndrome is often nonspecific, reflecting gastric outlet obstruction, which is frequently incomplete and includes symptoms such as nausea, vomiting and epigastric pain. A pre-existing history of symptomatic biliary tract disease can often be elicited (7). Dehydration and electrolyte abnor-

malities result from vomiting. Affected patients are usually elderly, and approximately 80% have concomitant, sometimes debilitating chronic diseases, thus increasing their risk for surgery (3,8).

Upper gastrointestinal hemorrhage in patients with Bouveret's syndrome has been reported in only five patients, originating from duodenal ulceration in four (1,8-10) and from an eroded cystic artery in one (6). Perforation of the duodenum by a large gallstone has also been described (11).

Although the diagnosis of Bouveret's syndrome is often established only intraoperatively, several radiological signs may suggest it. These include air or radiographic contrast in the biliary tree, demonstration of a stone on plain x-ray or upper gastrointestinal series, change in position of a previously known gallbladder stone and a dilated gas-filled stomach (12). The diagnosis of Bouveret's syndrome was established recently by computed tomography scanning in two cases (12,13).

Upper endoscopy has greatly facilitated the preoperative recognition of gastric outlet obstruction due to a large gallstone (14-22), and several authors have attempted endoscopic removal of the stone but were unsuccessful due to its

size (6,19,22,23). There is only a single case report from Italy where endoscopic extraction was accomplished (15). In another report, abdominal surgery was avoided by breaking up a duodenal gallstone using ultrasound-guided extracorporeal shock-wave lithotripsy before endoscopic extraction of the fragments (23).

In our patient, a large stone was found in the first part of the duodenum. Following its endoscopic removal, the fistula between the duodenum and the gallbladder was identified. A retrograde cholangiogram confirmed the fistulous connection as well as the absence of choledocholithiasis.

Because the patient remains asymptomatic and there was no intrabiliary air seen on a follow-up ultrasound, we assume that his fistula has closed. Being a high risk surgical patient, we elected to follow this patient and he currently remains asymptomatic.

The treatment for Bouveret's syndrome usually consists of surgical removal of the ectopic gallstone by enterolithotomy followed by fistula repair and cholecystectomy (1). However, as demonstrated in this report, endoscopic stone extraction is possible in selected cases, thus providing a valid alternative therapy for elderly, chronically debilitated patients.

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