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The Bouveret Syndrome: An Unusual Cause of Hematemesis

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The Bouveret Syndrome: An Unusual Cause of Hematemesis

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Gallstones are usually silent. Less commonly, patients with cholelithiasis develop symptoms and/or complications; biliary fistula occurs in 3% to 5% of the cases. When a large stone is passed and occludes the duodenum, gastric outlet obstruction (the Bouveret syndrome) may result. In reported cases, the stones are usually larger than 2.5 cm. The usual presenting symptoms are those of bowel obstruction: abdominal pain, nausea, and vomiting. Less commonly, the patients experience melena and, rarely, hematemesis. We describe a patient who had the largest stone reported to cause hematemesis rather than bowel obstruction and to be diagnosed endoscopically. The 5 X 4 X 3 cm stone was extracted surgically. Endoscopic diagnosis and extraction of stones up to 3 cm in size has been reported, avoiding the need for surgery. (Henry Ford Hosp Med J 1990;38:52-4)

Case Report

A 76-year-old white male who was receiving gold therapy for rheumatoid arthritis presented with a three-day history of nausea and vomiting of "coffee ground" material. He had mild nonradiating epigastric pain and a feeling of fullness, but no fever, chills, or jaundice. He gave no history of peptic ulcer, cholelithiasis, or liver disease. His vital signs were normal. The abdomen was soft with no palpable masses or organomegaly, and bowel sounds were present. There was slight mid epigastric tenderness. The stool was hemocult positive.

Hemoglobin was 127 g/L (12.7 g/dL), hematocrit was 0.358 (35.8%), and WBC count was $9.2 \times 10^9/L$ ($9,200/\mu L$) with normal differential count. Serum electrolytes were normal, BUN 15.7 mmol/L (44 mg/dL), and creatinine 141 $\mu\text{mol/L}$ (1.6 mg/dL). Total protein was 62 g/L (6.2 g/dL), albumin 32 g/L (3.2 g/dL), total bilirubin 10 $\mu\text{mol/L}$ (0.6 mg/dL), SGOT 18 IU/L, SGPT 19 IU/L, and triglycerides 0.95 mmol/L (84 mg/dL). Prothrombin time was 14 seconds.

Esophagogastroduodenoscopy (EGD) revealed a moderate amount of "coffee ground" material in the fundus of the stomach. A punched-out, ulcerated lesion seen in the postbulbar region (Fig 1) was highly suggestive of choledocystoduodenal fistula, and a large bile-stained stone was seen in the second portion of the duodenum (Fig 2). Attempts to remove and/or crush the stone endoscopically were unsuccessful because of its exceptionally large size.

Plain abdominal x-rays showed a large amount of gas in the colon and multiple calcified densities in the right upper abdomen which represented the stone seen endoscopically in the duodenum (Fig 3). Surgical exploration of the peritoneal cavity revealed a dense inflammatory reaction around the gallbladder and multiple adhesions encasing the duodenum. Biliary exploration and duodenostomy were performed with removal of the 5 X 4 X 3 cm tan-brown to black calculus (Fig 4). The patient's postoperative course was uneventful.

Discussion

Although gallstones are usually asymptomatic, patients may develop symptoms and/or complications of cholelithiasis such

as abdominal pain, nausea, and vomiting. Biliary fistula occurs in 3% to 5% of cases, and the most common sites of fistulous communication are between the gallbladder and duodenum

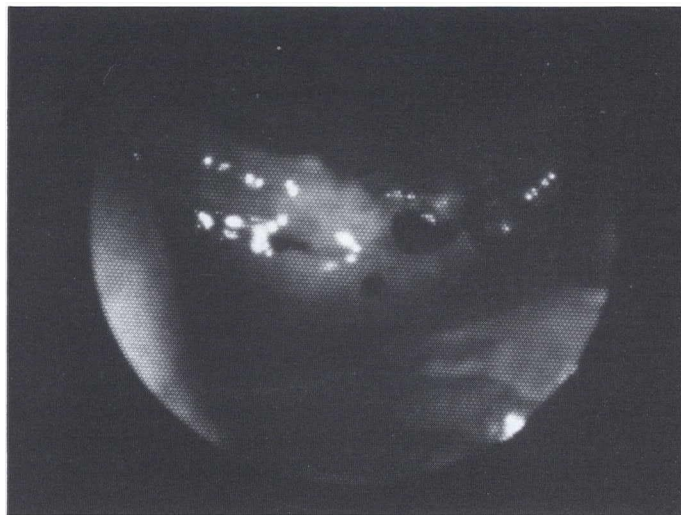


Fig 1—Punched-out, ulcerated area representing a choledocystoduodenal fistula seen endoscopically.

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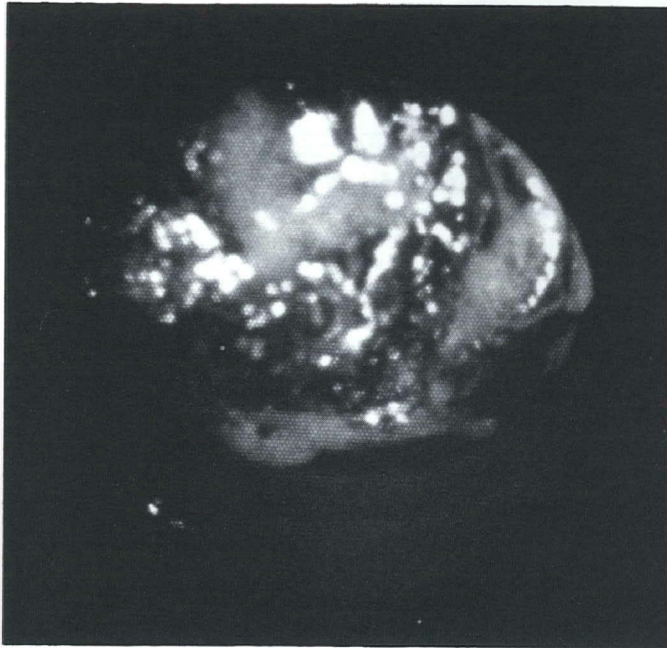


Fig 2—Large bile-stained stone in the second portion of the duodenum seen endoscopically.

(70%), the colon (14%), or the stomach (6%) (1-3). Choledochoduodenal fistula, identified in 4% of patients with fistulas, is usually due to duodenal ulceration rather than to biliary calculi (1-3). The usual presentation of gallstone ileus has the clinical features of intestinal obstruction with the radiographic finding of air in the biliary tree. The most common sites of intestinal obstruction by gallstones are the ileocecal valve (54% to 65%), the jejunum (27%), the duodenum (1% to 3%), and the colon (< 1%) (3-10).

Gastric outlet obstruction, an unusual complication of large gallstones, is termed the Bouveret syndrome. This rare entity is manifested by the presence of a cholecystoduodenal or choledochoduodenal fistula with a large gallstone (usually more than 2.5 cm) in the duodenal bulb causing gastric outlet obstruction. Since this syndrome was first described in 1770 by Beausier (subsequently by Bouveret in 1886), fewer than 90 cases have been reported (9). Females outnumber males in most reported cases by as much as 14 to 1 (9). Symptoms of the Bouveret syndrome, which are generally nonspecific, include epigastric pain, bloating, abdominal cramps, nausea, and vomiting which may be projectile (7,11). Apart from the acute presentation, most patients acknowledge having had biliary dyspepsia for several years. Gastrointestinal hemorrhage, a rare occurrence in the Bouveret syndrome, results from ulceration and erosion of the pyloric channel and the duodenum by the stone (12).

Preoperative diagnosis of the Bouveret syndrome is difficult. In most reported cases the diagnosis was made at laparotomy (Table). However, the clinical presentation and abdominal x-rays may suggest the diagnosis of biliary enteric syndrome. Radiologic signs of the Bouveret syndrome described by Rigler et al (13) included: 1) air in the biliary tree (93% of their cases); 2)

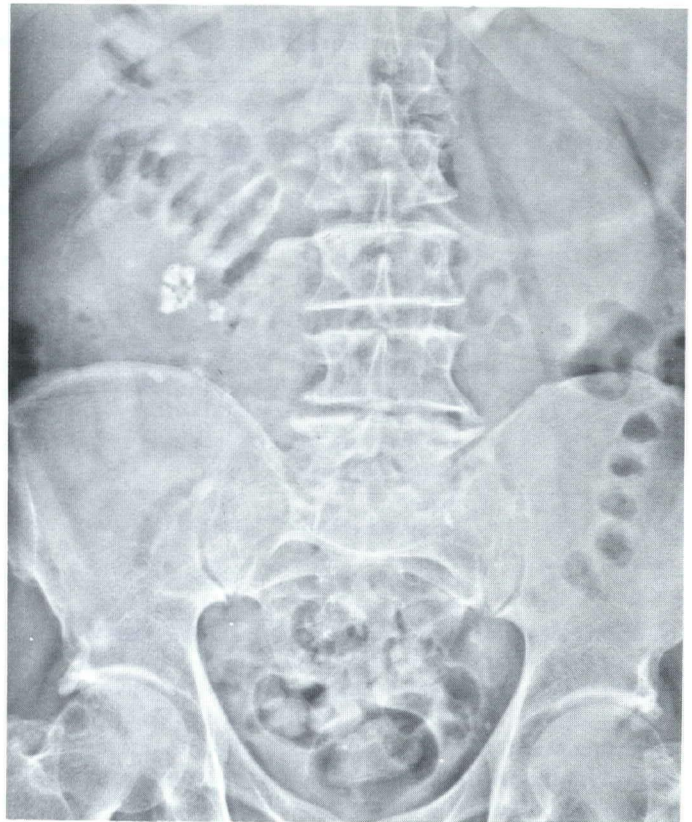


Fig 3—Abdominal x-ray showing the calcified area in the large gallstone.



Fig 4—The sectioned 5 X 4 X 3 cm tan-brown calculus after extraction.

dilated loops of small intestine which indicate ileus; 3) radiopaque calculi, seen either in the gallbladder, common bile duct, or small intestine; and 4) changes in the position of the radiopaque calculus, noted if serial x-rays are available (9,13,14). A

Table
Review of Reported Cases of the Bouveret Syndrome (1974 to 1988)

Author	Year	Number of cases	Hematemesis	Mode of Diagnosis	Size of Stone	Treatment
Mendelow (13)	1974	2	No	Laparotomy	Not reported	Surgery
			Yes	Laparotomy	Not reported	Surgery
Torgerson et al (19) Ayub & Michalko (16)	1979	1	No	Barium study	3 X 3 cm	Surgery
Bedogni et al (15)	1982	1	No	EGD	6 X 6 X 3 cm	Surgery
Patel et al (11)	1985	1	No	UGI series	Not reported	Endoscopy
				UGI series	8.5 X 4.5 X 3.5 cm	Surgery
Ryska et al (18)	1985	1	Yes	EGD	3 X 3 cm	Surgery
Hjelmqvist et al (20)	1985	3	Yes	Laparotomy	Not reported	Surgery
			Yes	Laparotomy	5 X 8 cm	Surgery
			Yes	Laparotomy	Not reported	Surgery
Chait & Lerner (12)	1986	1	Yes	Laparotomy	5 cm	Surgery
Ruiz (5)	1986	1	No	CT of abdomen	3 cm	Surgery
Maglinte et al (10)	1987	1	No	US, then UGI series	3 X 2.5 X 1.5 cm	Surgery
Godiwala et al (9)	1988	1	No	CT of abdomen	2 cm	Surgery
Current case	1990	1	Yes	EGD	5 X 4 X 3 cm	Surgery

EGD = esophagogastroduodenoscopy, UGI = upper gastrointestinal, CT = computed tomography, and US = ultrasound.

few cases of the Bouveret syndrome have been diagnosed preoperatively by ultrasound (7,8,10), computed tomography of the abdomen (5,6), or upper gastrointestinal radiographs (7,11,15,16). Few cases have been diagnosed by EGD (12,17-20), although this procedure has the potential of being a therapeutic as well as a diagnostic tool in some cases.

The current report describes the largest stone producing the Bouveret syndrome which presented mainly as upper gastrointestinal bleeding rather than bowel obstruction and was diagnosed initially by EGD (Table). Attempts to extract the stone endoscopically were not successful because of its exceptionally large size. Stones 3 cm or less in size have been extracted endoscopically by others (15,16), sparing the patient the need for surgical intervention. For this reason, we recommend that EGD be considered whenever the Bouveret syndrome is the suspected diagnosis.

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