Successful Endoscopic Management of Bouveret's Syndrome in a Patient with Cholecystoduodenocolic Fistulae

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Key Words

Bouveret · Gallstone · Cholecystoduodenal · Cholecystocolic

Abstract

Bouveret's syndrome, first described in 1896 by Léon Bouveret, is rare, limited to approximately 200 published case reports to date [Ariche et al.: Scand J Gastroenterol 2000;35:781–783]. It is a subgroup of gallstone ileus in which a cholecystoduodenal fistula allows the passage of a gallstone that obstructs the duodenum, causing gastric outlet obstruction. This case is unique as it describes Bouveret's syndrome in a patient with combined cholecystoduodenocolic fistulae. Gastric outlet obstruction was successfully managed endoscopically with lithotripsy. Both fistulae were subsequently managed conservatively without any complications.

Case Report

A 76-year-old lady presented with a one-week history of epigastric pain with radiation to the right upper quadrant. For the previous 24 h she had experienced repeated vomiting of black coffee ground material. She was haemodynamically stable but tender in her epigastrium without any guarding or rebound tenderness. She had a history of stable angina and was also noted to be a strict Jehovah's Witness.

Her full blood count was normal with a haemoglobin of 14.2 g/dl, urea and electrolytes slightly deranged with urea 7.8 mmol/l and K 3.3 mmol/l. Liver function tests were within normal range and C-reactive protein was elevated at 51 mg/l. A provisional diagnosis of upper gastrointestinal bleed was made and an urgent upper gastrointestinal endoscopy was arranged.

At upper gastrointestinal endoscopy there was a huge gallstone evident in the duodenal bulb ([fig. 3](#)). A through-the-scope balloon was passed distal to the stone and inflated. The balloon and the gallstone were then pulled back partially through the pylorus but it was not possible to dislodge the stone fully and the procedure was abandoned. A plain abdominal X-ray showed a radio-opaque shadow in the

right upper quadrant and pneumobilia ([fig. 1](#)). A decision to repeat the endoscopy in favour of emergency surgical intervention was made after discussion with the patient.

At repeat endoscopy, the gallstone was found in the body of the stomach ([fig. 4](#)), having migrated retrogradely following the previous procedure. An endoscopic lithotripter was then used to crush the stone into fragments of a suitable size for passage through the gastrointestinal tract.

Subsequent CT scan with contrast revealed pneumobilia and also a thick-walled gallbladder with a direct tract, with contrast communicating between the gallbladder and the first part of the duodenum representing a cholecystoduodenal fistula ([fig. 2](#)). In addition, there was a further tract of contrast between the gallbladder and the transverse colon representing a cholecystocolic fistula.

After endoscopic therapy the patient's symptoms resolved and she recommenced a normal diet without any complications. She was then discharged and reviewed as an outpatient where she remained asymptomatic.

Discussion

Léon Bouveret, the renowned French authority on gastric diseases, reported in 1896 the first two cases of gastric outlet obstruction due to gallstones [1, 2]. Bouveret's syndrome is usually the result of a cholecystoduodenal fistula. Although biliary-enteric fistulae are not uncommon, both Bouveret's syndrome and cholecystoduodenocolic fistulae are rare and limited to case reports [3]. Even more rare, as described here, is Bouveret's syndrome as the result of a cholecystoduodenocolic fistula.

The majority of patients with cholecystoenteric fistulae are elderly and there is a female preponderance (6:1) [4]. Cholecystoduodenal fistulae may cause gastric outlet obstruction as described or simply be asymptomatic. Cholecystocolic fistulae are usually asymptomatic with a benign clinical course. Diarrhoea is the most common presenting symptom and the typical clinical features of gallbladder disease are absent. The diarrhoea may be due to colonic irritation by bile acids, jejunitis caused by faecal reflux into the small intestine and steatorrhoea caused by lack of bile. Cholangitis has also been reported [5].

The largest review of Bouveret's syndrome encompassed 128 published cases [4]. This revealed that upper gastrointestinal endoscopy identified gastric outlet obstruction in virtually all cases, but the obstructing stone was identified in only 69%. Furthermore their review identified only 10 cases who were managed successfully by endoscopic therapy and only 3 cases by endoscopic lithotripsy. The majority of cases were managed surgically, usually by laparotomy.

If, as in this case, the obstructing gallstone can be removed by endoscopic therapy, further definitive surgical intervention can be deferred until the patient has fully recovered from gastric outlet obstruction. However, concomitant surgery to the cholecystenteric fistula is often discouraged on the basis that these complications in this elderly group of patients are rare [6].

Furthermore, reports indicate that biliary-enteric fistulas may close spontaneously after passage of the stone [7, 8], especially if the cystic duct is patent and residual gallstones are not present [9].

Fig. 1. Abdominal X-ray – evident pneumobilia.

Fig. 2. CT scan – evident cholecystocolic fistula.

Fig. 3. Gallstone in D1.

Fig. 4. Gallstone in fundus.

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