Unusual presentation of more common disease/injury

An unusual presentation of cholecystoduodenal fistula: vomiting of gallstones

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Summary

Bouveret's syndrome is defined as gastric outlet obstruction secondary to an impacted gallstone in the duodenum via a cholecystoduodenal or cholecystogastric fistula. Common radiological findings include pneumobilia, calcified right upper quadrant mass, pyloric or duodenal obstruction and cholecystoduodenal fistula. Initial attempts through endoscopic retrieval may be successful; however, results can vary. Surgical options include enterolithotomy or gastrotomy with or without cholecystectomy and fistula repair. We describe a unique case of Bouveret's syndrome with short-lived obstruction followed by vomiting of gallstones in a morbidly obese patient and discuss the complexities of investigation and management of these patients.

BACKGROUND

Cholelithiasis is a common health condition and only few patients (around 1–3%) develop rare complications such as choledochoduodenal fistulas. The pathogenesis of fistula formation is thought to be secondary to adhesions between a chronically inflamed gall bladder and an adjacent lumen, impeding its arterial blood supply and venous drainage.¹ Erosion through the bowel wall most commonly results in a gallstone ileus, most commonly obstructing the distal ileum where the lumen is at its narrowest.¹ In Bouveret's syndrome, the clinical picture is distinct, presenting with proximal gastrointestinal obstruction secondary to gastric or duodenal fistula.^{2 3}

Management can be complex, as the majority of these patients are elderly with multiple comorbidities and therefore death rates can be high. In obese patients, management can be challenging. We describe a case of cholecystoduodenal fistula presenting with short-lived obstruction followed by vomiting of gallstones in a morbidly obese patient and discuss the complexities of investigation and management in these patients.

CASE PRESENTATION

A 64-year-old lady was admitted with a 10 h history of severe, colicky epigastric and right upper quadrant (RUQ) pain radiating through to her back. Several hours later, she developed nausea and had one episode of vomiting. The contents of her vomit included two large gallstones (figure 1). She had no urinary or bowel symptoms and had otherwise been well prior to this. Her medical history included hypertension and hypercholesterolaemia for which she was on Metoprolol and Simvastatin, respectively. She was a non-smoker and had minimal alcohol intake.

Four years prior to this presentation, she had been admitted with biliary colic and deranged liver function tests (LFT's) and ultrasound confirmed cholelithiasis. Due to her large body habitus (body mass index >40), surgery was

postponed until she had lost a sufficient amount of weight. She had no further attacks within this 4-year period.

INVESTIGATIONS

On examination, she was in severe pain requiring morphine, afebrile and haemodynamically stable. She was morbidly obese, weighing 165 kg. Her abdomen was soft with tenderness in the epigastrium and RUQ. Inflammatory markers were within normal limits. She had a normal amylase of 27 and deranged liver function test's (LFT): bilirubin 55, alanine transaminase (ALT) 94, alkaline phosphatase (Alk P) 189 and gamma-glutamyl transferase (GGT) 222. Erect chest x-ray was normal with no evidence of free air under the diaphragm. Abdominal x-ray was unremarkable with a normal gas pattern and no evidence of pneumobilia. A CT was performed on day three of admission which revealed a cholecystoduodenal fistula with resulting pneumobilia and two ectopic gallstones both measuring 16 mm, in the mid-descending colon (figures 2A,B and 3). The gall bladder contained multiple gallstones but there was no evidence of choledocholithiasis. As the gastric outlet obstruction was short lived and spontaneously resolved through vomiting of the gallstones, there was no evidence of persistent gastric outlet obstruction on the CT. A nasogastric tube had also been placed temporarily in the interim.

DIFFERENTIAL DIAGNOSIS OF GASTRIC OUTLET OBSTRUCTION

- Foreign bodies
- Fibrotic ulcer
- Neoplasia
- Diverticulae

OUTCOME AND FOLLOW-UP

She was started on intravenous antibiotics and a decision made to intervene only if she developed obvious signs of cholangitis. On day four post-admission, she deteriorated

Figure 1 Two gallstones contained in patient's vomitus, the largest measuring 17 mm.

clinically and developed atrial fibrillation and a low blood pressure refractory to intravenous fluid resuscitation. She was transferred to the high dependency unit (HDU) where she received treatment for sepsis, presumed secondary to cholangitis, with inotropic support following which she made an uncomplicated recovery. As CT had confirmed the aetiology and she clinically improved following her HDU stay, endoscopic intervention was not required. She has remained symptom free for 11 months and has been referred to an obesity clinic for dietary support before being considered for surgical intervention.

DISCUSSION

Bouveret's syndrome was first described by Beaussier in 1770 but was subsequently named after Leon Bouveret when he published two case reports on this condition in the *Revue de Medicin* in 1896 (2). It tends to occur more commonly in women (65%) with a median age of 74.1 years.³ Due to the rarity and non-specific clinical symptoms and signs, the diagnosis can often be overlooked.⁴ There are several unique manifestations of the syndrome reported in the literature. The most recent comprehensive literature review included 128 cases by Cappell *et al*.

The most common presenting features include nausea and vomiting, abdominal pain, haematemesis, recent weight loss and anorexia. Clinically, the patients have abdominal distension and tenderness, signs of dehydration and fever.³

Figure 3 Abdominal CT: two gallstones in the mid-descending colon.

The major complications of this syndrome are intestinal obstruction and haemorrhage. Typically, an insidious presentation with lack of specific signs of biliary disease leads to a delay in diagnosis and resulting high death rate.⁴ In this case study, the patient presented with an acute history and short-lived gastric outlet obstruction. The unusual feature here was the retrograde migration of gallstones from the duodenum to the stomach, which would be a more plausible outcome in a patient having undergone previous gastric surgery, for example, a pyloroplasty.

Figure 2 (A) Abdominal CT (axial): cholecystoduodenal fistula and resulting pneumobilia (GB: Gallbladder, D: Duodenum). (B) Abdominal CT (coronal): cholecystoduodenal fistula and resulting pneumobilia. (C) Suboptimal ultrasound with poor appreciation of pneumobilia.

Common radiological findings include pneumobilia, calcified right upper quadrant mass or gallstone, gastric distension, dilated bowel loops, filling defects or mass in the duodenum, duodenal or pyloric obstruction and cholecystoduodenal fistula.³ Rigler's triad (small bowel obstruction, ectopic gallstones to the right of T12/L1 and pneumobilia) may be identified on both plain radiographs and CT images.⁴ The prevalence of obesity is increasing and therefore radiology departments are facing the repeated challenges of achieving good image quality.⁵ Difficulty lies in both the choice and interpretation of imaging and also the challenges in patient transport and accommodating large patients on imaging equipment.⁵

Sonography is limited due to poor penetration of the ultrasound (US) beam through the thick body parts and also due to the increased attenuation as it passes through the subcutaneous tissues and intraperitoneal fat.⁵ This is illustrated in figure 2C, where there is suboptimal imaging with poor appreciation of pneumobilia.

The limitations of CT include increased noise due to inadequate beam penetration and reduced field of view resulting in artefact and poor image quality due to cropping. In figures 2A,B and 3, beam hardening artefact is visible on the edges of these CT images.

Esoophagogastroduodenoscopy has been shown to be diagnostic in 69% of cases, the remaining patients with stones not visible but obvious mechanical obstruction.³ In these cases, it is possible that the gallstone is deeply embedded within the mucosa. US and CT are the preferred non-invasive tests to confirm the diagnosis and are better at delineating the gastroduodenal anatomy and visualising fistulas.³ However, approximately 15–25% of gallstones are isoattenuating and not well visualised on CT and in such cases magnetic cholangiopancreatography may be more useful.¹

Management is most commonly by enterolithotomy or gastrotomy with or without cholecystectomy and fistula repair. These have a high-success rate with low morbidity and mortality³ but this may not be appropriate in the morbidly obese patient and a conservative approach may be most suitable. The first endoscopic retrieval was successfully performed in 1985 by Bedogni *et al.* Endoscopic retrieval has a relatively low-success rate but with more innovative endoscopic techniques created, success rates are

much higher now and it is useful as an alternative therapy for patients not fit for surgery.¹ Alternative treatments include endoscopic laser, mechanic lithotripsy or extracorporeal shock-wave lithotripsy; however, these are limited by the need for multiple treatments and risk of converting a proximal gallstone ileus into a distal gallstone ileus by fragmentation of the stone.¹ In most cases, fistula repair is deemed unnecessary because the fistula may spontaneously close especially if no residual gallstones are present and the cystic duct remains patent.^{3,4}

Learning points

- ▶ Bouveret's syndrome is a rare consequence of gallstone disease and management in the obese patient can be complex and challenging.
- ▶ Imaging modalities should be chosen wisely and interpreted with caution.
- ▶ In this case, initial management was conservative, as the obstruction had spontaneously resolved through vomiting.
- ▶ In morbidly obese patients, we would advocate first-line treatment with endoscopic retrieval followed by encouraging weight loss.
- ▶ Other treatment options include gastric bypass with or without cholecystectomy and fistula repair.

Competing interests None.

Patient consent Obtained.

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