A case report and review of the literature of Bouveret syndrome

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ABSTRACT

Bouveret syndrome is a rare variant of gallstone ileus causing gastric outlet obstruction. It results from the formation of either a cholecystoduodenal or a cholecystogastric fistula and subsequent migration of gallstone into the duodenum or pylorus of stomach, causing obstruction. The first case was reported by Leon Bouveret in 1896. We report a case illustrating the rarity and severity of this condition, together with a review of the literature of the different methods of endoscopic and surgical treatment.

KEYWORDS

Bouveret syndrome – Gallstone disease – General surgery – Education – Gastric outlet obstruction

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Background

Bouveret syndrome is a rare cause of gastric outlet obstruction, representing less than 3% of cases of gallstone ileus.¹ It is an acquired cholecystoenteric fistula between the gallbladder and the duodenum. The formation of the fistula is caused by chronic inflammation, with an increase in the intraluminal pressure, directly causing wall ischaemia and perforation allowing gallstone passage into the bowel.² We report a case of Bouveret syndrome, illustrating the rarity and severity of this condition with a review of the literature on the different methods of endoscopic and surgical treatment.

Case history

An 89-year-old man with a past medical history of atrial fibrillation, hypertension and gastro-oesophageal reflux disease presented to the emergency department with acute confusion, coffee-grounds vomiting and anorexia. On examination, he had a distended, tender abdomen with localised guarding in the epigastrium. He had a white blood cell count of $13 \times 10^9/l$ (normal range $4\text{--}10 \times 10^9/l$) and C-reactive protein of 43 mg/dl (normal range 0.8–3.0 mg/dl). An initial diagnosis of upper gastrointestinal bleeding led to the patient having an oesophagogastroduodenoscopy, which demonstrated an erythematous oesophagus and a swollen pylorus with a large obstructing stone (Fig 1). After a second endoscopic attempt to remove the stone failed, the patient was referred to the emergency surgical team. Computed tomography demonstrated a 3.5

cm gallstone lying in the second part of duodenum, which extended proximally through the pylorus leading to gastric outlet obstruction (Fig 2).

At surgery, an upper-midline mini-laparotomy and longitudinal pyloromyotomy was made to extract multiple large gallstones (Fig 3) aided by Desjardins forceps. The pylorus was closed transversely primarily with 3.0 polydioxanone interrupted extramucosal sutures followed by an omental patch. An abdominal drain was placed intraoperatively, with an uneventful postoperative period.

The patient was treated with total parenteral nutrition support, proton pump inhibitor and prophylactic intravenous broad-spectrum antibiotics. He was managed in intensive care for the first 24 hours. On day five, oral nutrition was commenced after a normal Gastrografin swallow study. He was discharged on the seventh day from his admission date and was reviewed in the outpatient clinic with no further concerns.

Discussion

Bouveret syndrome is a rare variant of gallstone ileus causing gastric outlet obstruction. The common risk factors are being elderly with an average age of 70 years, female sex and gallstones greater than 2.5 cm, which have higher chances of impaction and subsequent bowel obstruction.^{4,5} The symptoms are nausea and vomiting (85%), abdominal pain (70%), haematemesis, weight loss and anorexia. Clinical signs are obstructive in nature, such as abdominal tenderness and distension with altered bowel habits. Findings suggestive of Bouveret syndrome include Rigler's

triad: a dilated stomach, pneumobilia and a radio-opaque shadow suggesting an enteric gallstone.⁶ The mainstay of management is surgical intervention to remove the

Figure 1 Endoscopic view of large gallstone obstructing the pylorus.

Figure 2 There is a large calcified gallstone lying within the pylorus of the stomach, measuring 3.5 cm. The proximal stomach is fluid filled but not significantly dilated. There is a further calculus lying lateral to the pylorus which lies within the cholecystoenteric fistula. There is associated pneumobilia and gas within the gallbladder.

Figure 3 Extraction of multiple large gallstones.

obstructing gallstone. Non-operative management is associated with a mortality of 26.5%.⁷

Non-operative management (endoscopic options)

Bedogni *et al* described the first successful case of endoscopic treatment of Bouveret syndrome in 1985.⁸ Endoscopic visualisation of the stone is possible in only about 69% of cases, owing to submucosal embedding of the stone.⁹ The endoscopic findings suggestive of Bouveret syndrome are dilated stomach containing old digested food from gastrointestinal obstruction, together with a hard, non-fleshy and convex mass at the obstruction.⁵ Multiple endoscopic modalities have been used for treatment of Bouveret syndrome including endoscopic stone removal with nets or baskets.⁶ For stones that cannot be removed using nets or baskets, lithotripsy techniques such as mechanical, electrohydraulic, intracorporeal laser, and extracorporeal shockwave therapy can be used to break the stone prior to endoscopic extraction. After lithotripsy, all fragments of the stone must be removed to prevent a postoperative gallstone ileus.² Risks include intestinal wall haemorrhage or perforation.¹⁰ Note that 42% of surgical patients have already failed an endoscopic trial for gallstone removal.^{11,12}

Operative management

Surgery is required in about 90% of cases, with mortality ranging from 19% to 24%.⁵ Within the current literature, there are three surgical approaches. A common surgical approach consists of laparoscopic or open gastrotomy, pyloromyotomy or duodenotomy at or immediately proximal to the site of obstruction.^{15,14} If it is possible to manoeuvre an impacted duodenal gallstone into the stomach a gastrotomy can be applied for stone removal.¹⁵ When the gallstone is present in an area that may be difficult to access during open surgery, endoscopy can be used as an adjunct to help mobilise the stone into a more favourable location for performing enterotomy.⁶

Table 1 An overview of existing evidence and their respective management strategies and outcomes.

Study	Type	Management strategies and outcomes
Caldwell <i>et al</i> (2018) ⁶	Case report and literature review	Site: 6 × 4 cm gallstone in gastric antrum. Procedure: laparoscopy; open cholecystectomy + partial gastrectomy + Billroth II gastrojejunostomy + intraoperative cholangiogram. Outcome: 1) first-line option, endoscopic retrieval ± use of adjuncts; 2) laparoscopic extraction of gallstone through an enterotomy; 3) stone extraction + cholecystectomy + fistula repair.
Bhattarai <i>et al</i> (2016) ¹⁴	Case report	Site: first part of the duodenum; Procedure: laparotomy + anterior pyloromyotomy + stone extraction.
Mir S, <i>et al</i> (2015) ¹⁶	Systematic review of 113 cases of recurrent gallstone ileus reported in 91 articles	Management: 1) enterolithotomy (86.7%); 2) enterotomy with cholecystectomy and repair of fistula. Outcome: mortality in the past 25 years associated with: enterolithotomy (4.8%), single stage surgery (22%).
Qasaimeh <i>et al</i> (2014) ¹	Case report and literature review	Site: 3.5 × 5 cm gallstone in the first part of the duodenum. Procedure: laparotomy + anterior longitudinal duodenotomy + stone extraction. Outcome: 1) simple stone extraction related mortality rate 12%; 2) definitive one-stage procedure-related mortality rate 20–30%.
Keller <i>et al</i> (2014) ¹³	Case report and literature review	Site: duodenal bulb. Procedure: laparotomy + gastrotomy + stone extraction. Management: 1) endoscopic retrieval; 2) enterolithotomy.
Mavroeidis <i>et al</i> (2013) ¹²	Case report and literature review	Site: 5.7 × 3.8 × 4 cm gallstone in D2 + D3. Procedure: laparotomy + Kocher's manoeuvre + jejunotomy + stone extraction. Management: 1) endoscopy-assisted retrieval; 2) simple enterolithotomy; 3) one-stage procedure. Outcome: simple duodenotomy related mortality 12%; combined treatment-related mortality 20–30%.
Doycheva <i>et al</i> (2009) ¹¹	Case report and literature review	Site: 3.3 cm gallstone in the second part of the duodenum. Procedure: endoscopic retrieval using various devices including a retrieval net, snare, stone extracting basket, mechanical lithotripter and lithotripsy basket. Management: 1) initial treatment option endoscopic retrieval; 2) simple enterolithotomy; 3) combined procedure. Outcome: 1) simple duodenotomy related mortality 12%; 2) combined treatment-related mortality 20–30%.
Park <i>et al</i> (2009) ¹⁰	Case report	Site: cystic lesion of the first and second parts of the duodenal wall (containing multiple gallstones) causing gastric outlet obstruction. Procedure: laparotomy + cholecystectomy + excision of a portion of the duodenal wall.
Ayantunde <i>et al</i> (2007) ²	Retrospective review of 22 cases	Management: 1) 20 patients had enterolithotomy; 2) 2 patients had a one-stage procedure. Outcome: 5 perioperative deaths, 1 episode of cholangitis 18 months after enterolithotomy alone.
Cappell <i>et al</i> (2006) ⁵	Literature review of 128 cases	Outcome: endoscopy revealed gastroduodenal obstruction in nearly all cases but identified the obstructing stone in only 69% cases. The following endoscopic findings are suggestive of Bouveret syndrome: a dilated stomach containing old digested food from gastrointestinal obstruction together with a hard and non-fleshy mass at the obstruction.

Table 1 (continued)

Lowe <i>et al</i> (2005) ³	Literature review of 44 cases from 39 papers	Management: 1) 91% of patients underwent surgery (64% laparoscopy/open enterolithotomy, 36% one-stage repair); 2) 9% of patients had nonsurgical treatment. Outcome: overall mortality 12–24%.
Reisner <i>et al</i> (1994) ⁷	Literature review of 1001 cases	Management: 1) simple enterolithotomy + stone extraction; 2) one-stage procedure. Outcome: enterolithotomy-associated mortality 11.7%; one-stage procedure-associated mortality 16.9%.

Many authors have commonly used the ‘open gastrotomy’ approach as described in our case. Laparoscopic enterolithotomy and gastrotomy have been reported and are safe and effective in the management of this condition. It is important to examine the remaining small intestine to look for any residual stones that may cause postoperative gallstone ileus. If there are no residual stones in the gallbladder with a patent cystic duct, theoretically the simple stone extraction method can lead to the fistula closing spontaneously.^{6,9}

The second approach of surgery is more invasive and continues to be actively debated on whether patients should have a ‘one-stage procedure’ that involves a gastrotomy or enterotomy and stone extraction, with or without cholecystectomy and repair of cholecystoduodenal fistula. This may require partial resection of the duodenum and common bile duct surgery with tertiary centre involvement. Mir *et al* reports in the past 25 years, enterotomy alone as the treatment for recurrent gallstone ileus was associated with mortality of 4.8%. Whereas surgery involving enterotomy with cholecystectomy and repair of cholecystoenteric fistula had a mortality rate of 22.2%.¹⁶ The third approach is a ‘two-stage procedure’ involves enterotomy with stone extraction followed by an elective cholecystectomy and repair of fistula, once the inflammatory process around the gallbladder improved. The benefits include preventing recurrent cholecystitis, cholangitis, gallstone ileus and the occurrence of fistulising gallbladder carcinoma.^{6,9} Chowbey *et al* illustrated the success of the laparoscopic endostapling for patients with a cholecystoenteric fistula; however, the mean age of the patients was 57.2 years with no comment on comorbidities.¹⁷ Reisner *et al* reviewed 1001 reported cases of gallstones ileus and concluded a one-stage procedure carried a mortality of 16.9%, compared with 11.7% for simple enterolithotomy.⁷ Table 1 gives an overview of existing evidence and their respective management strategies and outcomes.

There is a general consensus between surgeons that less is better when treating patients with Bouveret syndrome.¹⁸ Historically, the one-stage approach was found to be associated with a high rate of postoperative morbidity and mortality; hence most surgeons advocate the enterolithotomy-only approach.¹⁹ However, our review highlights a lack of empirical evidence to support one type of surgery over another. Owing to the rarity of the condition, the data are scarce on primary repair compared

with spontaneous closure of the fistula and the true benefits of performing cholecystectomy.

Conclusion

A high degree of suspicion is required for diagnosis of Bouveret syndrome and it may be diagnosed incidentally on initial imaging workup for gastric outlet obstruction. Although there is no standardised protocol for its treatment, endoscopy is a useful initial diagnostic and potential therapeutic adjunct. In the case of impacted stones, assistant options such as lithotripsy can be used for fragmentation of the stone and endoscopic retrieval. Owing to the high failure rate of endoscopy, surgery remains the mainstay treatment modality. Laparoscopic or open extraction of the stone through a gastrotomy or duodenotomy is the treatment of choice. Enterotomy alone is the preferred option in elderly patients with multiple comorbidities. Cholecystectomy and fistula repair as a single- or a two-stage procedure can be considered after careful planning.

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