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A Stone and a Net: A Case Report of Bouveret's Syndrome Managed Endoscopically

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Abstract

Bouveret's syndrome refers to gastric outlet obstruction secondary to (an) ectopic gall stone(s) in the gastric pylorus or duodenum. Despite its non-specific symptoms, it is a potentially fatal surgical emergency. As it is unlikely to resolve with conservative management, prompt diagnosis of Bouveret's syndrome is key to improving patient outcomes. In this case report, we discuss an 82-year-old man who presented with the complaints of feeling unwell and vomiting for three days. He was initially treated for a chest infection until the sixth day of admission, when he underwent a computed tomography scan, which showed an obstructing gallstone in the anterior pylorus. He was thereafter referred to our team. Following the advice of the regional hepatobiliary surgery team, we requested an oesophagogastroduodenoscopy (OGD) before a planned surgical intervention. Fortunately, the obstructing gallstone was easily retrieved using an endoscopic net during the OGD, and the patient subsequently recovered rapidly. This is one of the very few cases in the medical literature where an obstructing gallstone was retrieved intact with an endoscopic net, without the need for lithotripsy. This case serves as a reminder that, in carefully selected patients with Bouveret's syndrome and appropriately sized stones, endoscopic stone retrieval may represent a minimally invasive therapeutic option.

Categories: General Surgery, Gastroenterology
Keywords: bouveret's syndrome, case report, gallstone, gastric outlet obstruction, oesophagogastroduodenoscopy

Introduction

Gallstones are common in many of the world's major population subgroups [1]. In the United Kingdom, while the prevalence of gallstones increases with age, affected individuals are often asymptomatic [2]. Symptomatic gallstones, termed gallstone disease, may manifest as biliary colic, cholecystitis, cholangitis, or pancreatitis [3]. Rarely, gallstones may cause obstruction outside the biliary tree, including in the gastric pylorus [4], duodenum [5], small bowel [6], or large bowel [7]. This usually occurs in the context of chronic inflammation and the formation of a cholecysto-enteric or choledocho-enteric fistula [8], allowing one or more gallstones to migrate into the lumen of the bowel and cause mechanical obstruction.

Bouveret's syndrome (BS), which refers to gastric outlet obstruction secondary to ectopic pyloric or duodenal gallstones, is a rare form of mechanical gastric outlet obstruction. First described in two postmortem patients by Bonnett in 1841, the first preoperative diagnosis was made by French physician Leon August Bouveret in 1896, giving rise to the eponymous term [9]. The rarity of the condition has made the development of consensus management guidelines difficult. In this case report, we describe the clinical course and management of BS in an 82-year-old man who presented atypically. This case report has been written in line with the SCARE 2020 guideline [10]. Consent for the clinical data to be published was obtained from the patient and documented.

Case Presentation

An 82-year-old man with a background of hypertension, type 2 diabetes, and compensated heart failure was reviewed at home by his general practitioner on account of feeling unwell and vomiting for three days. On initial examination, he was clinically dehydrated, tachypnoeic (respiratory rate: 26 cycles per minute), with low oxygen saturation (92% on breathing room air), and a low-grade fever (38°C). Bedside investigations showed hyperglycaemia (13 mmol/L), ketonuria (+++), and glycosuria (+++). Suspecting diabetic ketoacidosis, the general practitioner promptly referred the patient to our emergency department, where he was initially assessed by a medical intern. The intern noted that he had a new cough, but the patient reported no change in his bowel habit. Clinical observations in the emergency department showed a normal respiratory rate (20 cycles per minute) and temperature (36.1°C), while oxygen saturation was 97% on supplemental oxygen (4 L/minute). On examination, there were crepitations in both lung bases. His abdomen was soft and non-tender, and bowel sounds were present. There was no lower limb oedema, and both calves were non-tender. Point-of-care testing showed hyperglycaemia (14.9 mmol/L), ketonaemia (blood ketone: 2.9 mmol/L), and a hypokalaemic metabolic alkalosis (pH: 7.46, bicarbonate: 35 mmol/L, and base excess: 9.51 mmol/L). Lactate was normal (1.7 mmol/L). The urine pH was 5.5 with negative nitrites and leucocytes. The ketonaemia subsequently normalised with intravenous rehydration. Initial laboratory

examinations showed leucocytosis (22 × 10^9/L), neutrophilia (19.7 × 10^9/L), and elevated C-reactive protein (182 mg/L). Blood biochemistry showed hypokalaemia (2.9 mmol/L), hypochloraemia (88 mmol/L), and elevated blood urea (9.8 mmol/L) but normal creatinine. Liver function tests were normal apart from mildly elevated bilirubin of 32 μmol/L. His chest radiograph showed atelectasis at the right lung base but no pneumoperitoneum, while the abdominal radiograph (Figure *1*) revealed a distended stomach but no dilated bowel loops. There were also two faint radiopaque bodies in the right upper abdominal quadrant, but these were not noticed at the time. The initial diagnosis was lower respiratory tract infection. The patient was admitted under the acute medical team and managed with intravenous fluids, potassium replacement, antimicrobial therapy, and anti-emetics. An abdominal ultrasound scan was also requested.

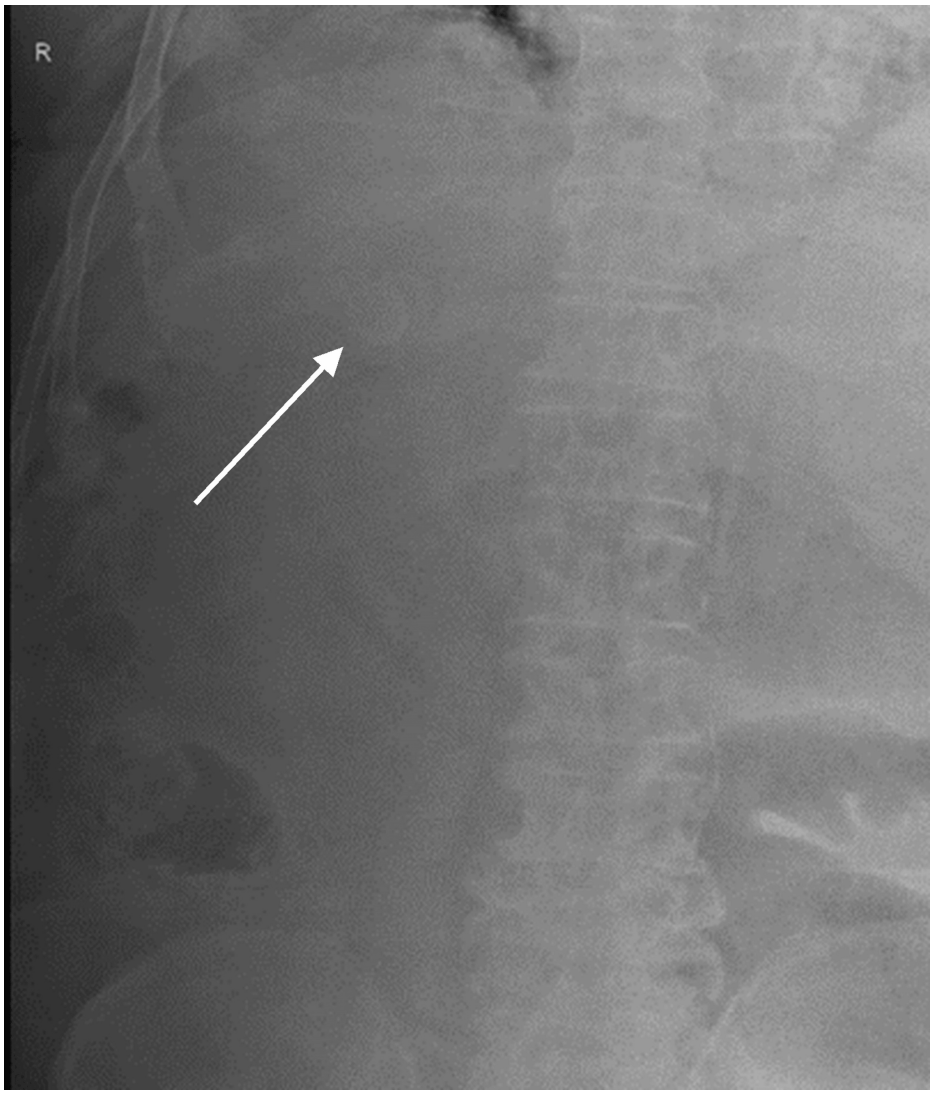


FIGURE 1: Patient's abdominal radiograph on the day of admission. Two faint, round radiopaque materials can be seen in the right upper quadrant (arrow).

The patient was reviewed by a specialty doctor in acute medicine on the second day of admission. The management plan remained essentially the same. The patient underwent an abdominal ultrasound on the third day of admission. The scan showed a dilated common bile duct measuring 12 mm, but the gallbladder was not visualised. The patient underwent a repeat liver function test on the same day, which was normal except for a raised gamma-glutamyl transferase (103 U/L). The patient's ultrasound scan finding was discussed with the gastroenterology team, who advised magnetic resonance cholangiopancreatography (MRCP). However, the MRCP was not completed until the patient was discharged from the hospital. Meanwhile, the patient's CRP continued to rise despite intravenous antibiotics, reaching a peak of 217 mg/L on the third day of admission. In addition, his abdomen became progressively distended, and his vomiting did not settle. However, he continued to open his bowel. He therefore underwent a computed tomography (CT) scan of his abdomen with intravenous contrast on the sixth day of admission. The CT scan (Figure *2*) showed gastric outlet obstruction with an ectopic gallstone in the anterior aspect of the pylorus and a

possible cholecysto-pyloric fistula. Given the CT scan findings, a referral was sent to our team (general surgery).



FIGURE 2: An axial view of the computed tomography scan (with intravenous contrast) of the patient's abdomen on the sixth day of admission. Prominent findings include a distended stomach with an air-fluid level (arrowhead), a gallstone impacted in the pylorus (white arrow), and a second gallstone (black arrow), possibly in the gallbladder wall. The gallstone in the pylorus measured 17 mm on the tomograph. Pneumobilia is not obvious on this view.

At initial review by the senior house officer and registrar in general surgery, the patient reported no abdominal pain. On examination, his abdomen was distended but non-tender. He subsequently had a nasogastric tube inserted and was kept on intravenous fluids with oral sips of water, but with no food allowed. Repeat blood tests done on the day showed a reduced CRP (48 mg/L) with normal white cell count and electrolyte profile, except for raised bicarbonate (30 mmol/L). Following review by the surgical consultant on the seventh day of admission, the patient underwent a gastrografin swallow and follow-through radiography with abdominal radiographs taken one, two, and five hours post-gastrografin (Figure *3*). The abdominal radiographs revealed that much of the contrast was held up in the stomach up to five hours post-contrast ingestion. The patient was subsequently discussed with the regional hepatobiliary surgery team who, on reviewing the CT scan images, felt that the pyloric stone was rather small to cause gastric outlet obstruction and recommended an oesophagogastroduodenoscopy (OGD) to directly visualise the stomach and exclude other pathologies before any surgical intervention.

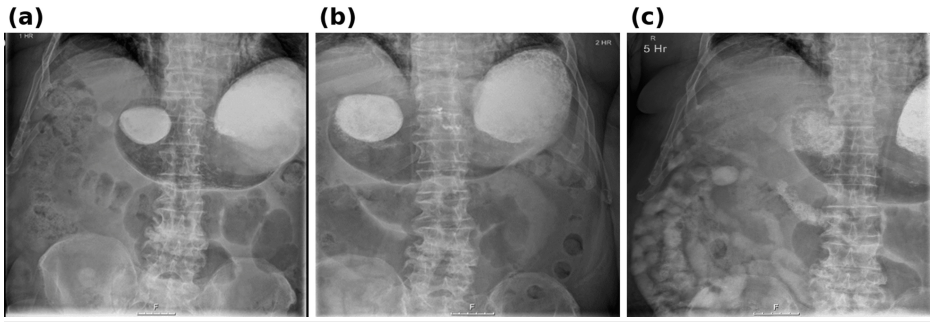


FIGURE 3: Abdominal radiographs (a) one hour, (b) two hours, and (c) five hours post-gastrografin meal showing that most of the ingested contrast remained in the stomach.

As advised by the regional hepatobiliary surgery team, the patient underwent an OGD on the eighth day of admission under local anaesthetic with an overtube for airway protection. At OGD (Figure *4*), a 2 cm stone was found at the pylorus causing gastric outlet obstruction. In addition, a suspected submucosal lesion was noted along the greater curvature of the stomach. The stone was removed with an endoscopic net, and the duodenum was visualised following stone removal. The duodenum was inflamed, but no fistula tract was visualised. The stone was lamellated and had the typical appearance of a cholesterol stone (Figure *5*). The nasogastric tube was not reinserted after the OGD.

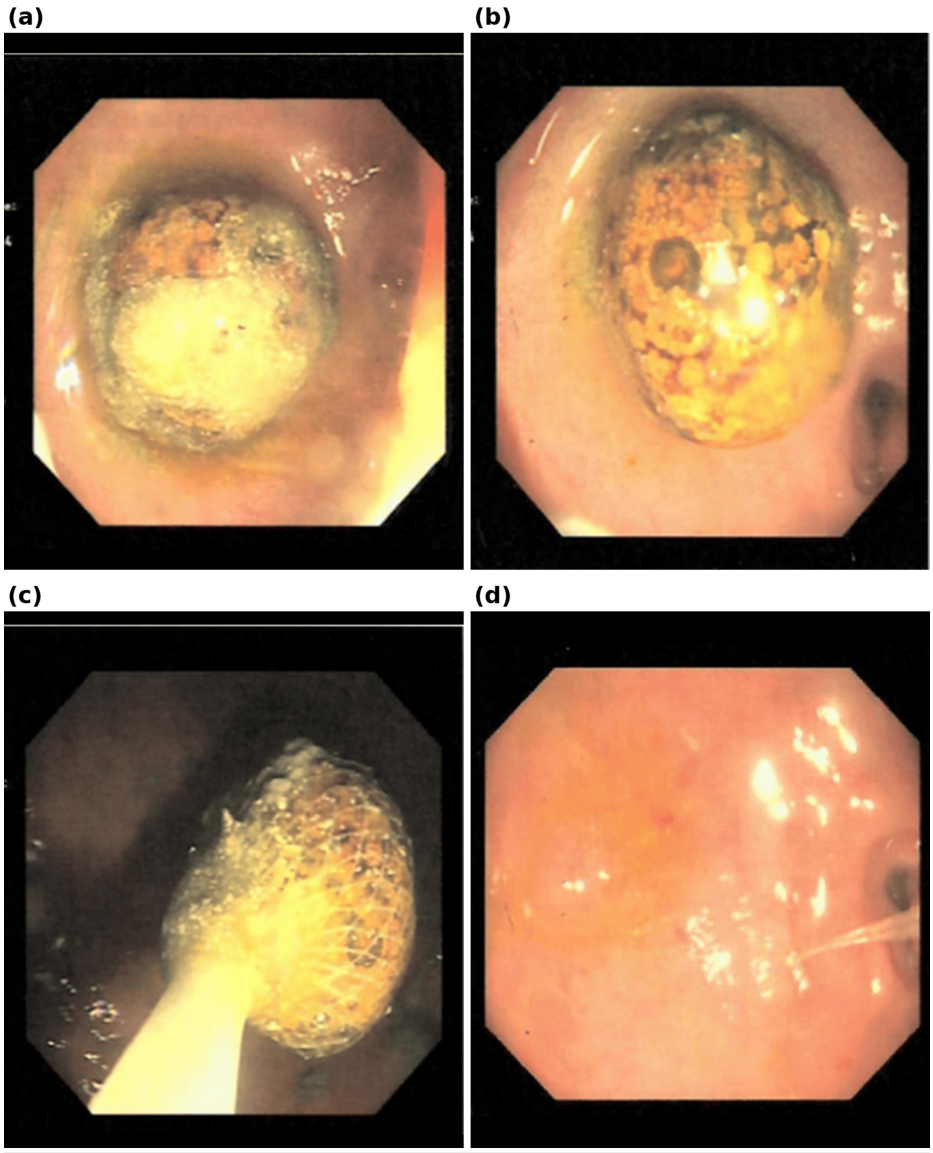


FIGURE 4: Endoscopic views of (a, b) a gallstone lodged in the gastric pylorus causing gastric outlet obstruction, (c) stone retrieval with an endoscopic net, and (d) gastric pylorus after stone retrieval.



FIGURE 5: The retrieved gallstone shows a lamellated appearance.

Following his return to the ward from the endoscopy department, the patient was allowed to take oral fluids, which he tolerated. He subsequently progressed to a light diet after some hours, and resumed a normal diet the following day. He had no further episodes of vomiting and his abdominal distension gradually resolved. The patient remained well over the subsequent few days, and he was declared fit for discharge on the 10th day of admission. He remained in the hospital for six more days while waiting for social care arrangements. He was discharged home with lifelong apixaban for paroxysmal atrial fibrillation detected during the admission and eight weeks of high-dose proton pump inhibitor. In addition, he was booked for outpatient endoscopic ultrasound to investigate the submucosal lesion noted at OGD. Given his age and clinical frailty, there were no plans for additional surgical intervention if he remained well. However, he was booked for follow-up in the general surgery clinic. The patient was satisfied with his overall hospital experience. He was particularly pleased that he did not have to undergo open surgery to remove the obstructing gallstone.

The patient had an endoscopic ultrasound scan done four weeks after his discharge. The scan showed three gallstones in his common bile duct. He subsequently underwent endoscopic retrograde cholangiopancreatography to clear the common bile duct stones. He was contacted by telephone six weeks after his discharge; he remained well and was tolerating a normal diet.

Discussion

This case highlights the non-specific presentation of BS. This likely contributed to our patient's delayed diagnosis. Given its rarity, clinicians often consider other diagnoses in patients presenting with the symptoms of nausea, vomiting, epigastric pain, and abdominal distension. Nevertheless, as highlighted in this case, certain radiographic and laboratory findings, in conjunction with the relevant clinical presentation, should prompt clinicians to consider BS as one of the possible diagnoses when assessing patients. In our patient, the initial point-of-care tests showed hypokalaemic metabolic alkalosis with paradoxical aciduria, which is a hallmark of gastric outlet obstruction [11]. Rigler's triad, which includes ectopic gallstone(s), pneumobilia, and features of intestinal obstruction, is a radiographic finding which may be observed in patients with BS and the related condition of gallstone ileus. In a review of the abdominal radiographs of 81 case reports of BS, Ferhatoğlu and Kartal noted the presence of a calcified gallstone in 37% of cases, while all three components of the triad were observed in 30-35% of cases [8]. Therefore, the presence of any abnormal opacities and calcifications on abdominal radiographs, as observed in our patient, should prompt a consideration of BS in any patient presenting with relevant symptoms.

The dramatic improvement in our patient's CRP on the sixth day of admission may be due to a combination of rehydration, intravenous antimicrobial therapy, and gastric decompression with a nasogastric tube. This highlights the fact that supportive measures, even in the absence of a definitive diagnosis, could significantly improve patients' physiological parameters, buying time for additional investigations.

Although there is currently no universally agreed gold-standard investigative modality for BS, most centres rely on one or more of abdominal CT scan, magnetic resonance imaging (MRI), and OGD to make an accurate diagnosis [12]. Each modality has its merits and demerits. CT scanning is widely available and quick, but it involves ionising radiation, while MRI, which is radiation-free, may not be suitable for individuals with

metallic implants. OGD is invasive, but it may be diagnostic and therapeutic, as our case highlights. Nevertheless, given its higher sensitivity compared to OGD, CT scan of the abdomen with the use of intravenous contrast is the favoured investigative modality for BS in most centres [12-14]. It is superior to plain abdominal radiographs in identifying Rigler's triad and may help with the planning of therapeutic interventions by identifying the fistula connection between the biliary tree and the stomach [12].

There is no consensus on the ideal management strategy for BS. Most surgeons agree that it is unlikely to resolve with conservative management. Endoscopic, laparoscopic, and open surgical approaches have been described to facilitate the retrieval of the obstructing gallstone(s). Endoscopic treatment for BS involves OGD and stone fragmentation using mechanical, laser, electrohydraulic therapy, and/or extracorporeal shockwave devices before retrieval of stone fragments [8,15]. Several attempts may be needed to successfully break the stone into smaller pieces that can be retrieved with an endoscopic basket [4]. Complications associated with this modality include aspiration of gastric content, damage to the gastric mucosa and surrounding tissues from the fragmentation process [4] and, rarely, gallstone ileus secondary to distal impaction of relatively large stone fragments [16]. In cases involving smaller gallstones with a favourable orientation, endoscopic stone retrieval with a basket or net may be all that is required to relieve the obstruction [17,18]. As demonstrated in our case, this approach may definitively address the obstruction, leading to rapid recovery without the exaggerated metabolic response associated with surgery. We recommend that it be considered in carefully selected patients.

In cases where endoscopic attempts have been unsuccessful, surgical intervention is unavoidable. The ideal surgical treatment for BS is still debated. However, given the frail and multimorbid status of most patients with BS, more extensive surgery may result in greater complication rates and poorer outcomes [19]. Therefore, many surgeons favour retrieving the obstructing stone through a gastrotomy, pylorotomy or duodenotomy, with cholecystectomy and fistula closure considered later as a staged procedure only if the patient's clinical condition and medical fitness allow for it [15]. Stone retrieval is usually done as an open procedure, although, in skilled hands, a laparoscopic approach may result in less morbidity and faster recovery [20]. Regardless of the surgical modality employed, it is important to examine the bowel in its entirety to ensure that there are no additional stones that may cause obstruction [4].

While leaving gallstones and an intact choledocho-duodenal fistula in situ increases the risk of cholangitis, BS, and gallstone ileus in the future [21], an interval cholecystectomy and fistula closure carries significant morbidity and mortality risks in frail patients [22]. Our patient's frailty and comorbidities, together with reported cases of good short-to-medium-term outcomes in patients who did not undergo cholecystectomy and fistula closure [22], ultimately justified adopting a conservative approach for managing his gallstones and fistula.

Conclusions

This case highlights the diagnostic and therapeutic dilemma commonly encountered in patients with BS. Due to its rarity and non-specific presenting symptoms and examination findings, the diagnosis of BS requires advanced imaging techniques, which tend to occur much later in the clinical course of the condition, thereby delaying treatment strategies. Endoscopic approaches, in particular OGD and stone retrieval, should be considered in carefully selected patients to reduce the morbidity and mortality associated with surgical intervention. The key to improving outcomes for patients with BS is early diagnosis, adequate resuscitation, and the selection of the treatment modality that minimises the stress response and hospital stay.

Additional Information

Author Contributions

All authors have reviewed the final version to be published and agreed to be accountable for all aspects of the work.

Concept and design: Olakunle G. Akanji, Kaviraasen Mootoosamy, Abdalazeez Ahmad

Acquisition, analysis, or interpretation of data: Olakunle G. Akanji, Kaviraasen Mootoosamy, Abdalazeez Ahmad

Drafting of the manuscript: Olakunle G. Akanji, Kaviraasen Mootoosamy, Abdalazeez Ahmad

Critical review of the manuscript for important intellectual content: Olakunle G. Akanji, Kaviraasen Mootoosamy, Abdalazeez Ahmad

Disclosures

Human subjects: Informed consent for treatment and open access publication was obtained or waived by all participants in this study. **Conflicts of interest:** In compliance with the ICMJE uniform disclosure form, all

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