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## Case Report

# Upper gastrointestinal bleeding revealing a Bouveret syndrome: A case report<sup>☆</sup>

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## ABSTRACT

Bouveret's syndrome is an uncommon cause of gastrointestinal obstruction. It's a result of the passage of a gallstone through a fistula connecting the gallbladder with the duodenum or stomach. The diagnosis is challenging due to its atypical clinical manifestations. There have been a few reported cases of Bouveret syndrome presenting with gastrointestinal bleeding. Treatment options include both endoscopic and surgical approaches. We present the case of a 92-year-old woman admitted to the emergency department for upper gastrointestinal bleeding. Gastroscopy revealed gastric stasis upstream of a calculus inducing an obstruction of the bulb. The computed tomography (CT) scan showed a cholecystoduodenal fistula with a calculus lodged in the bulb. The patient underwent a gastrostomy with extraction of the calculus. Postoperative course was uneventful and the patient was discharged home. In the majority of cases, Bouveret's syndrome is revealed by an upper gastrointestinal obstruction, but other signs, such as gastrointestinal bleeding, can be seen. The diagnosis is confirmed by an imaging method that highlights Rigler's triad. The management can be either endoscopic or surgical depending on the patient's general condition. The diagnosis is often difficult due to the lack of specificity in the symptoms. Presently, there exists no consensus concerning the appropriate approach for its management.

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## Introduction

Bouveret's syndrome is a rare complication of biliary lithiasis. It is a pyloric or duodenal obstruction by a gallstone that has migrated through a fistula between the biliary system and the duodenum or the distal part of the stomach. These fistulas are due to chronic inflammation and the development of adhesions between the bile ducts and the duodenum causing necrosis of the intestinal wall with the creation of a passageway for the stones [1]. The main manifestation is intestinal obstruction, but rare complications such as bleeding in the digestive tract may also occur. Treatment also presents challenges given the prevalence of this condition among the elderly population, who are at elevated surgical risk and often present complex treatment scenarios. As a result, it becomes crucial to familiarize oneself with the array of therapeutic modalities available. Our work aims to raise awareness among practitioners regarding uncommon origins of gastrointestinal bleeding, particularly when risk factors for such conditions are present.

## Case presentation

We report the case of a 92-year-old female patient with a medical history of type 2 diabetes under oral antidiabetic. She was admitted to the emergency department due to a one-month history of delayed food vomiting, accompanied by instances of hematemesis and melena. The patient did not report abdominal pain, jaundice, constipation, or fever. On physical examination, the patient's vital signs were normal: she patient was conscious, hemodynamically stable, with no jaundice and no fever. Abdominal examination revealed tenderness in the right hypochondrium with no Murphy's sign and no palpable mass.

Laboratory tests revealed a normal hemoglobin level at 12 g/dL, with a normal liver function test: total bilirubin at 6 mg/dL, aspartate aminotransferase (ASAT) at 6 UI/L, alanine aminotransferase (ALAT) at 11 UI/L, alkaline phosphatase (ALP) at 103 UI/L, albumin at 35 g/L, C-reactive protein (CRP) at 46 mg/L (normal range <10 mg/dL). Renal function and blood ion levels were found to be within normal ranges (Table 1).

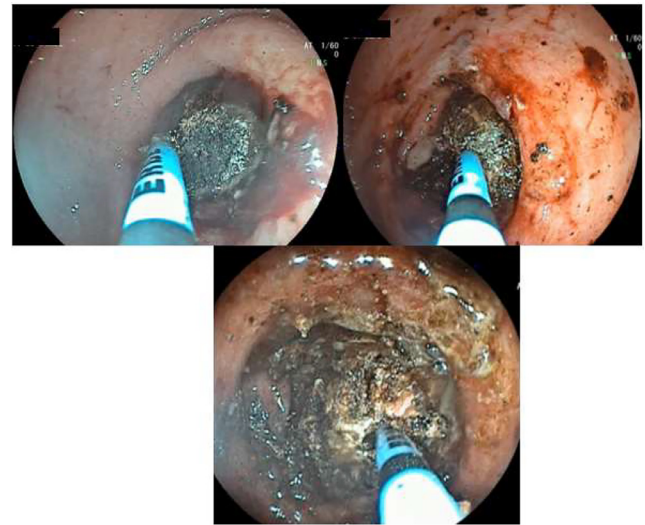
Gastroscopy revealed stasis located upstream from a sizable macrocalculus causing obstruction in the bulbar lumen. Additionally, esophagitis of grade D, as per the Los Angeles classification, was observed. Attempts at endoscopic fragmentation of the calculus proved unsuccessful (Fig. 1).

Abdominal ultrasonography revealed a Gallbladder multilithiasis with no dilation of the common bile duct. Abdominal CT scans revealed gastroduodenal distension, displaying a hydroaerobic level located upstream of an impacted stone within the duodenal bulb. The stone, measuring 4 cm in length, was accompanied by the presence of a cholecystoduodenal fistula. Additionally, air-filled pockets were identified within the common bile duct (Fig. 2).

The patient underwent a laparotomy procedure during which various adhesions were noted, and it was identified that a small gallbladder had formed a fistula with the initial section of the duodenum. A longitudinal anterior gastrotomy

**Table 1 – Overview of patient's laboratory tests.**

	Results	Normal range
Bilirubin	6	2-12 mg/L
AST	6	5-34 UI/L
ALT	11	0-55 UI/L
GGT	32	0-36 UI/L
ALP	103	<150 UI/L
CRP	46	0-5 mg/L
WBC	6860	4000-10,000/uL
HB	12	12-16 g/dL
Platelet Count	249,000	150,000-400,000/uL
K+	3	3.5-5 mmol/L
Na+	138	135-145 mEq/L
Creatinine	5	7-11 mg/L
Urea	0.25	0.15-0.45 g/L
Albumin	35	35-50 g/L



**Fig. 1 – Macrocalculus obstructing the pylorus with failed endoscopic extraction.**

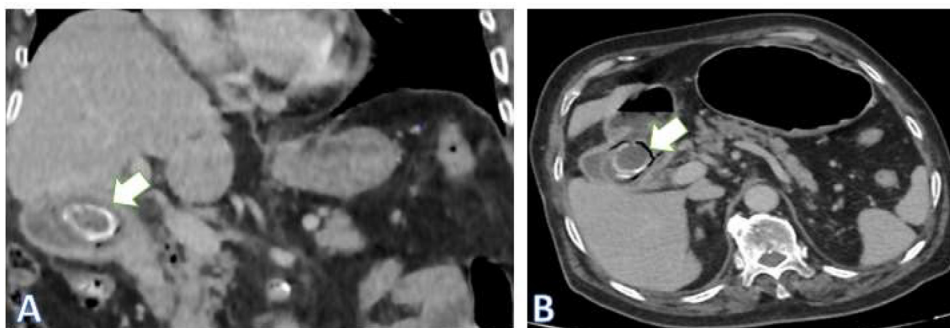
was conducted on the antrum, facilitating the extraction of the gallstone (Fig. 3). The fistula was not repaired during the operation.

Following the operation, the patient's recovery progressed without complications, leading to her discharge on the fourth day after the procedure.

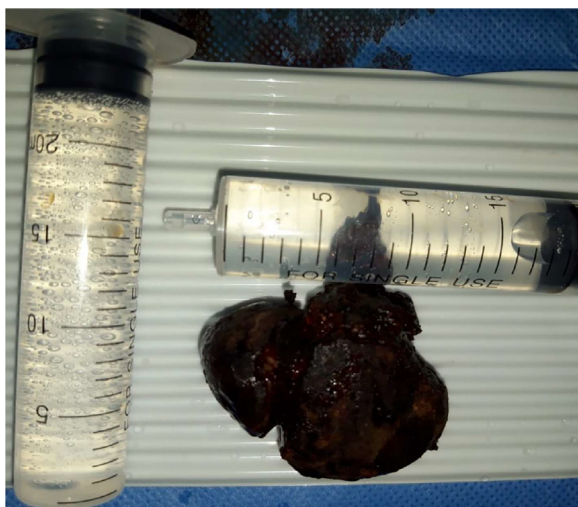
## Discussion

Due to the complexity of this case, we conducted a comprehensive literature review focusing on the diagnosis and management of this specific medical condition.

Bouveret's syndrome was first described in 1896 by Leon Bouveret [1]. It arises from the occurrence of biliodigestive fistulas, an exceedingly rare complication of biliary lithiasis, accounting for less than 1% of cases [2]. The formation of this fistula results from the process of pressure-induced necrosis



**Fig. 2 – Coronal (A) and transverse (B) sections depicting an obstructing calculus at the bulb with accompanying upstream hydroaerobic distention on CT.**



**Fig. 3 – Postoperative picture of the stone after extraction.**

impacting both the gallbladder wall and the adjoining organ (the duodenum or the stomach). This process often involves the development of adhesions between these organs, facilitated by chronic inflammation and the entrapment of a stone in the neck of the gallbladder [3].

Bouveret's syndrome is mostly seen in women, with the sex ratio female/male: 1.86, with an average age of 74 years [4]. Approximately 43%-68% of documented cases exhibit a medical history of hepatic colic, jaundice, or acute cholecystitis. However, it's important to recognize that Bouveret syndrome can also be the initial presentation of biliary lithiasis [5]. The mortality of Bouveret's syndrome ranges from 12% to 30%, as it occurs in the elderly with multiple comorbidities [6].

In the majority of cases, the symptoms and clinical signs are not very specific, which adds complexity to the diagnostic process. The main symptoms are: Nausea and vomiting (87%), abdominal pain (71%), hematemesis (15%), and weight loss (14%). Clinical examination may be unremarkable or reveal tenderness of the right hypochondrium [4]. Biological tests can reveal hyperleukocytosis, increased levels of inflammatory markers, especially CRP, and impaired liver function [3].

Concerning radiological assessment, the abdominal plain X-ray holds limited diagnostic significance. The identification of Rigler's triad (characterized by pneumobilia, intestinal obstruction, and the presence of a calcified ectopic stone within the digestive tract lumen) is considered pathognomonic for Bouveret's syndrome. However, this triad is observed in only 10%-50% of cases [7]. Abdominal ultrasound can be used to look for gallbladder stones, to detect thickening of the wall, or to identify bladder atrophy. Discontinuity of the bladder wall and the presence of air in the lumen may also be noted, indicating a biliodigestive fistula. Rigler's triad is detected in only 11% of cases [3].

CT scan can detect Rigler's triad in more than 80% of cases [8] and it allows the visualization of the obstructing stone, the level of obstruction, and the fistula site, but approximately 15%-25% of stones are radiolucent, meaning they might not be detectable through standard imaging [7]. The Magnetic Resonance Imaging (MRI) proves highly valuable for diagnostic purposes, as the triad can be identified in all cases (100%) [8]. Moreover, MRI facilitates the visualization of radiopaque stones that might not be discernible using CT scans [7].

Upper endoscopy can be performed, particularly in patients presenting with upper gastrointestinal tract bleeding (as observed in our patient) or chronic vomiting. The stone's visualization is attainable in around 69% of cases. Nevertheless, it's important to note that visualizing the fistula through this procedure is a rare occurrence [4].

In terms of treatment, the scarcity of this syndrome has led to a lack of consensus, resulting in ongoing debates between endoscopic and surgical approaches [8]. Most patients with Bouveret syndrome are elderly and often had multiple concurrent health conditions, which can potentially complicate surgical interventions. As a result, endoscopic treatment is preferred. There are various options, such as utilizing a basket for the extraction of small stones. However, this method can potentially impact the esophagus when the stone is large [9]. Additionally, there are other complications such as potential harm to the esophagus due to sharp-edged stones, stones migrating into the mediastinum, damage to the vocal cords, and even cardiac arrhythmia stemming from vagus nerve stimulation [10]. If extraction fails (size > 2.5 cm), other endoscopic options are available, including mechanical, hydroelectric, or



laser lithotripsy, which can fragment the stone to facilitate its extraction. However, it is associated with some risks, including obstruction of the lower bowel due to migration of stone fragments, the risk of bleeding, and perforation have also been reported [1]. The limitations of endoscopic treatment include the fact that the fistula cannot be closed, but this is not necessary in most cases [9]. The success rate of endoscopic treatment ranged from 37% to 43% in a systematic review by Ong [10]. Additional therapeutic approaches encompass lithotripsy utilizing extracorporeal shock waves, either as a standalone method or in conjunction with endoscopic treatment [11]. An initial successful instance of using extracorporeal lithotripsy for treating an impacted duodenal stone was reported by Holl et al. [9]. This method is used for large and well-defined stones. In most cases, an endoscopic extraction is required following the fragmentation of the stone.

If endoscopic treatment proves ineffective or is unfeasible due to the absence of suitable technical resources, surgical intervention becomes a viable consideration. Surgical options encompass performing a gastrotomy, pylorotomy, or duodenotomy, based on the stone's location. For the stones located in the jejunum or distal duodenum, an enterotomy is necessary accompanied by the examination of the small intestine to look for any potential additional stones that could potentially contribute to postoperative obstructions [9].

The topic of cholecystectomy along with fistula repair has sparked a debate. On one side, proponents recommend them at the same operative time to prevent later biliary complications, specifically cholecystitis, cholangitis, and gallbladder carcinoma [1]. And on the other side, those who propose against repairing fistulas, because there is a potential for spontaneous closure and the associated risk of harming adjacent structures due to the presence of substantial inflammation [11]. The mortality rates after cholecystectomy with fistula closure versus simple duodenotomy are approximately 20%-30% and 12%, respectively [2].

## Conclusion

Bouveret's syndrome is a rare complication of biliary lithiasis. The diagnosis is rarely made because of the nonspecific symptoms; it is based on radiologic elements consisting of the Rigler triad. There is no standard protocol for therapeutic management; endoscopy plays a diagnostic and therapeutic role, especially in patients in whom surgery is contraindicated. Surgical treatment consists of gastrotomy, duodenotomy, or enterotomy, depending on the location of the stone; cholecystectomy is not mandatory in these patients because of the surgical risk.

## Ethical approval

Not applicable

## Patient consent

Informed consent was obtained from the patient. The patient's identity has been anonymized, and any potentially identifying information has been removed.

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