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## **Bouveret's Syndrome: Diagnostic Dilemmas and Therapeutic Strategies - A Comprehensive Educational Approach for Healthcare Professionals**

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**Abstract:**

Bouveret's syndrome, a rare complication of gallstone disease, poses a significant diagnostic and therapeutic challenge, particularly in elderly individuals with multiple comorbidities. The mortality rate ranges from 12-30%, emphasizing the importance of prompt and accurate diagnosis. Nonspecific symptoms, such as nausea, vomiting, and abdominal pain, contribute to delayed diagnosis. Healthcare professionals, especially those involved in gastroenterology and surgery, require comprehensive education on Bouveret's syndrome. Training should emphasize the diverse diagnostic modalities, including the use of imaging techniques such as X-rays,

ultrasound, and MRI. Awareness of the syndrome's rarity and collaboration among specialists from various fields are crucial for effective management. Additionally, educational programs should underscore the need for individualized treatment plans, considering patient parameters such as age, comorbidities, and the inflammatory state of surrounding tissues. While endoscopic procedures exhibit lower success rates, they are often preferred due to the high risk associated with open surgical procedures in elderly patients. Continuous medical education should also highlight the potential link between diabetes and Bouveret's syndrome, emphasizing the importance of recognizing risk factors and ensuring timely medical intervention. In conclusion, enhancing the knowledge base of healthcare professionals through targeted education is essential for improving the diagnosis and management of Bouveret's syndrome, ultimately contributing to better patient outcomes.

**Aim of this study:**

Study of the etiology, epidemiology, pathophysiology, clinical presentation, diagnostic approaches, treatment strategies and complications of Bouveret syndrome based on the current literature. In order to update knowledge about this disease entity.

**Material and methods:**

A systematic review of the scientific and medical literature from the PubMed and Google Scholar databases was carried out.

**Key words:** Bouveret's Syndrome; gallstone ileus; cholecysto-duodenal fistula; Rigler's triad

**INTRODUCTION**

Bouveret's syndrome, named after French surgeon L. Bouveret, who first published an article about it in 1896, is a rare complication of gallstone disease (occurring in 0.3% to 0.5% of cases) leading to intestinal obstruction in the vicinity of the duodenum caused by gallstones. [1, 8, 9] The gallstone enters the GI tract, most often, through the cholecysto-duodenal fistula or, less commonly, through the cholecysto-gastric fistula. [1, 9, 12] As of 2016, only 315 cases of this syndrome had been reported. [1, 8, 11] The condition predominantly affects elderly individuals with multiple comorbidities, and its diagnosis is often delayed due to nonspecific symptoms,

resulting in a high mortality rate ranging from 12-30%. [1, 8, 10] The treatment of choice is endoscopy, followed by surgery if unsuccessful. [1, 11,18]

## **ETIOLOGY**

Bouveret's syndrome is the rarest type of gallstone ileus. It arises due to the passage of gallstones from the gallbladder through the bilioenteric fistula and the obstruction of the intestinal lumen by the stones, near the beginning of the duodenum. [5, 8 ,9] Recurrent cholecystitis stimulates necrosis of the gallbladder wall, which results in the formation of a cholecysto-intestinal fistula. [5, 8, 10] The process of necrosis is often accompanied by the formation of adhesions between the gallbladder and the stomach or duodenum. [15]

Scientifically proven risk factors for Bouveret's Syndrome include gallstone disease (especially large stones with a diameter of 2 to 8 cm), surgically altered gastrointestinal anatomy, female gender and age over 60. Moreover, 43% to 68% of patients have a history of recurrent biliary colic, jaundice or acute cholecystitis. [1, 8, 10]

Patients with Bouveret's syndrome, due to their age, often have multiple comorbidities. Numerous cases of Bouveret's syndrome have been documented in patients with diabetes in the literature. [22, 23, 24, 25].

## **EPIDEMIOLOGY**

Bouveret's syndrome is extremely rare, accounting for 1% to 3% of cases of ileus caused by gallstones [1, 8, 9] and 0.3% to 0.5% of complications of cholelithiasis. [1] Gallstones typically cause obstruction in the terminal ileum, and in order to induce obstruction in the region of the duodenum, they must attain considerable dimensions (generally at least 2.5 cm in diameter). Hence, Bouveret's syndrome is an exceedingly uncommon condition. [7]

## **PATHOPHYSIOLOGY**

As a result of recurrent inflammation of the gallbladder and possible adhesion of the gallbladder directly to the gastrointestinal wall, along with the mechanical pressure exerted by the stones, ischemia of the gallbladder and intestine walls may occur, resulting in the formation of a cholecysto-intestinal fistula. [1, 11, 21] Cases of fistula formation related to a neoplastic process

in the gallbladder have also been reported. [1, 18] The most common type of fistula is the cholecysto-duodenal fistula (68% of cases), while the cholecysto-gastric fistula is the rarest (5% of cases). [1, 6, 12]

## **SYMPTOMS**

In the diagnosis of Bouveret's Syndrome, it is crucial to pay special attention to patients with a history of gallstones and symptoms of gastric outlet obstruction. [2, 10, 11] Symptoms are usually nonspecific and most commonly include: nausea and vomiting (85% of patients), abdominal pain and bloating (70% of patients), upper abdominal and right upper quadrant pain, dehydration. [1, 3, 4] Rarely, bloody vomiting may occur due to erosions of the duodenal and visceral arteries or the expulsion of stones in vomitus. [1, 7, 10] Symptoms typically persist for 5-7 days before the patient seeks medical assistance. [1, 7]

The results of the physical examination are also nonspecific, with attention drawn to dry mucous membranes, abdominal bloating and tenderness, metallic sounds of intestinal peristalsis and generalized jaundice. [1, 3, 4]

## **DIAGNOSTICS**

Due to the rarity of this condition, there are no standardized guidelines for the diagnosis and treatment. [1, 2, 28] Bouveret's syndrome needs to be differentiated from peptic ulcer perforation, acute pancreatitis, gastric volvulus, cancer of the gastric outlet, internal hernia, gastric pyloric stenosis caused by bezoar and neoplastic fistula. [7, 10, 21] Laboratory test results are nonspecific and may indicate mechanical jaundice, including elevated liver enzymes and leukocytosis. [4, 7, 11] In patients presenting with symptoms of gastrointestinal obstruction, standing abdominal X-rays are routinely performed - typically revealing Rigler's triad (pathognomonic for Bouveret syndrome): small bowel obstruction, the presence of air in the bile ducts (pneumobilia) and ectopic gallstone. [2, 3, 8] This triad is visible on only 30% of X-ray images. [6, 17] Additionally, abdominal ultrasound (USG) can be conducted, which may reveal gallstones with possible inflammatory changes in the gallbladder and the presence of fluid around the gallbladder or identify a fistula. [2, 4, 9] Moreover, ultrasound can reveal stomach distension. [7, 10] X-ray of the gastrointestinal tract with oral administration of contrast can demonstrate signs of obstruction, the presence of gallstones in the gastrointestinal

tract, gastric distension, and, in rare cases, there may be contrast enhancement of the gallbladder or bile ducts, indicating the existence of a fistula connecting the biliary system directly to the gastrointestinal lumen. [2, 16, 21] CT scanning may yield similar results to the abdominal X-ray overview (potentially revealing Rigler's triad), but it is characterized by higher sensitivity, more frequently detecting the presence of a fistula, air in the biliary ducts, or an ectopic gallstone. [2, 4, 8] The most accurate diagnostic imaging modality is magnetic resonance imaging (MRI) as it allows for the identification of Rigler's triad in every patient with Bouveret's Syndrome. Additionally, MRI can visualize non-calcified stones that may not be apparent in computed tomography scans. [15] If a patient presents with bloody vomiting, the diagnostic procedure of choice is usually esophagogastroduodenoscopy, where gastrointestinal obstruction is almost always observed, but the gallstone is visualized in only about 2/3 of all cases. [2, 3] In 20-40% of all cases, the final diagnosis is only confirmed during surgery. [10]

## **TREATMENT**

Chances of spontaneous resolution of Bouveret's Syndrome after conservative treatment are minimal, and a displaced stone can lead to distal obstruction. [1] Treatment options include endoscopic, laparoscopic or laparotomy procedures. [2]

Considering that most patients with Bouveret's Syndrome are elderly with multiple comorbidities, various endoscopic interventions have been developed to reduce the mortality associated with open surgical procedures. [2, 8, 13] Before therapeutic endoscopy, the distended stomach should be decompressed with a nasogastric tube to reduce the risk of aspiration of food content. [1] Endoscopic methods include the use of endoscopic nets/baskets, laser lithotripsy, mechanical lithotripsy, and extracorporeal shock wave lithotripsy, with an efficacy ranging from 10-25%. [2, 3, 8] Using an endoscopic basket allows the removal of small stones, as larger stones may damage the esophagus. Additionally, the use of a basket to remove a sharp stone can cause esophageal damage and lead to stone migration into the mediastinum or damage to the vocal cords. [15] Endoscopic procedures can also complement each other; for example, if a stone is too large for mechanical removal, lithotripsy can be applied first to break it into smaller pieces, which can then be removed. [2, 3] After lithotripsy, it is crucial to remove all resulting gallstone fragments, as they may cause distal ileus. [3, 8, 10] The most common complication associated with endoscopy is gastrointestinal wall bleeding or perforation. [3, 10, 11] Unfortunately, endoscopic procedures exhibit a significantly lower success rate compared to

surgical interventions (43% versus 94.1%). [1] Endoscopy may be limited by the unavailability of specialized equipment or the inability to visualize the gallstone. [2, 28] The primary cause of endoscopic procedure failure is often attributed to the excessively large size of the gallstone. [6, 10] Nevertheless, surgery should be preceded by either endoscopic or laparoscopic intervention to reduce the likelihood of postoperative complications. [4, 10, 20]

About 91% of patients undergoing endoscopic treatment subsequently require surgery. [5, 28, 29] Three methods of surgery are described in the literature. The first involves gastrotomy, pylorotomy, or duodenotomy with gallstone removal. If the gallstone is challenging to access in open surgery, endoscopy is used as a complementary approach to reposition the stone. [28] It is crucial to examine the entire small intestine to detect all stones, as stones left in the intestine may later cause distal intestinal obstruction. The second surgical method is the "single-stage procedure," which includes gastrotomy or enterotomy with stone removal, repair of the gallbladder-duodenal fistula, and potential cholecystectomy. [3, 27] While this procedure may seem ideal, only a few patients with Bouveret's Syndrome are able to undergo such an invasive and high-risk surgery. [26] This approach is associated with higher mortality and a greater risk of postoperative complications compared to enterotomy. [3, 8, 26] The third and final surgical method is the "two-stage approach," involving enterotomy initially, followed by cholecystectomy and repair of the cholecysto-duodenal fistula once the inflammatory condition around the gallbladder decreases. [3, 8, 27]

The benefits of cholecystectomy and fistula repair are questionable, as patients with Bouveret's Syndrome are typically elderly and present with multiple comorbidities. Cholecystectomy is recommended if the patient is younger or if there is a risk of gallbladder cancer. [6] Recent advancements in treatment include minimally invasive surgical procedures using robotic assistance. The utilization of robots in surgery allows for greater precision and is less burdensome for the patient. Robotic-assisted procedures are performed at lower intra-abdominal pressure compared to typical laparoscopic surgery, which is advantageous for patients with cardiovascular diseases. [5] The combination of endoscopic methods and open surgery results in a 20–30% higher mortality rate compared to a simple duodenotomy. [14]

Due to the rarity of this condition, there are no precise treatment guidelines for Bouveret's Syndrome. Therefore, collaboration among physicians from various specialties is essential, along with the development of an individualized treatment plan for each patient. [1, 5, 28] Prompt removal of the obstructing stone is crucial, as intestinal obstruction significantly increases mortality. [14] The therapeutic strategy should take into account multiple parameters,

including the patient's overall condition, any comorbidities, age, inflammatory state of surrounding tissues, location of the obstruction, size of the fistula and stones, as well as the number of gallstones. [19, 27]

## **COMPLICATIONS**

Complications may also depend on the chosen treatment method. Incomplete lithotripsy can result in recurrent intestinal obstruction and the use of shock waves lithotripsy may damage surrounding tissues. Failure to close the fistula can lead to disease recurrence and acute pancreatitis. Surgical intervention, after unsuccessful endoscopic treatment, carries the risk of bleeding or infection. Cholecystectomy may unintentionally damage the bile ducts, especially in cases of current inflammation. Untreated Bouveret's Syndrome can lead to malnutrition due to gastric outlet obstruction. The most dangerous complication of gallstone obstruction is intestinal perforation. [1]

## **CONCLUSIONS**

Bouveret's syndrome is a rare complication of gallstone disease, causing intestinal obstruction near the duodenum. It predominantly affects elderly individuals with multiple comorbidities, leading to a mortality rate ranging from 12-30%. Diagnosis is challenging due to nonspecific symptoms, including nausea, vomiting, and abdominal pain. Treatment options include endoscopy and surgery, with endoscopic procedures exhibiting lower success rates. Bouveret's Syndrome poses a diagnostic challenge due to its rarity, and prompt, individualized treatment considering patient parameters is crucial to mitigate the associated high mortality rate. The collaboration among specialists is essential for effective management. It is noteworthy that a significant portion of patients developing Bouveret's Syndrome have chronic diabetes. We hypothesize that diabetes, due to polyneuropathy and reduced pain perception, may also be a risk factor for the occurrence of this syndrome. Patients experience recurring episodes of acute gallbladder inflammation leading to fistula formation, which in individuals with diabetes induces significantly diminished pain sensations, and patients may not seek appropriate medical assistance during such episodes.



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## **Data Availability Statement**

As a review paper, our work does not present new data or analyses. Therefore, there are no specific datasets or data availability to report. The information and findings presented in this review are based on previously published studies, which can be accessed through their respective sources as cited in the reference section.

## **Conflict of Interest Statement**

The authors declare that there are no significant conflicts of interest associated with this research work.

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