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CASE REPORT

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Early onset Bouveret syndrome: a unique case in a young patient

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Abstract

Background Bouveret syndrome is a rare etiology of gastric outlet obstruction, presenting with clinical manifestations that resemble those of several gastric pathologies. Timely diagnosis is imperative to mitigate potentially fatal consequences associated with this condition.

Case presentation This case report involves a male patient, aged 35, who exhibited symptoms of intense abdominal discomfort, nausea, and vomiting over a 24-h period. The individual experienced identical symptoms of lesser intensity on three separate occasions throughout the preceding 4-month period, during which they resorted to analgesic medication for temporary relief. A contrast-enhanced computed tomography (CT) scan of the abdomen was performed, which identified the presence of the Rigler triad, characterized by pneumobilia, a cholecysto-antral fistula, and an ectopic gallstone lodged in the gastric antrum resulting in an obstruction. The patient's diagnosis of Bouveret syndrome was determined based on the imaging findings. A surgical procedure known as cholecystectomy was conducted, which confirmed the existence of the sizable cholecystoduodenal fistula that was previously detected in the computed tomography (CT) scan. The surgical procedures that were conducted included a partial duodenectomy, a gastrojejunostomy, a cholecystectomy, and a common bile duct exploration with the removal of common bile duct stones. After examining the bile duct, a t-tube was inserted into the bile duct.

Conclusion This case demonstrates a distinctive occurrence of a condition in a young man, typically observed in elderly females. There is a lack of established treatment guidelines for this particular entity. The appropriate course of action depends on various factors, including the patient's condition, CT scan results, medical history, presence of co-existing illnesses, and intraoperative circumstances. These considerations are taken into account when determining whether a single-step surgical strategy or a multi-step approach should be pursued in order to minimize complications in patients. In this case, due to favorable conditions, a single-step approach was taken that yielded promising results.

Keywords Syndrome, Outlet obstruction, Gall stones

Background

Bouveret syndrome is a rare and potentially life-threatening condition characterized by the presence of a gallstone in the stomach or duodenum, causing an obstruction in the gastrointestinal tract. This obstruction can lead to symptoms such as severe abdominal pain, nausea, vomiting, and jaundice depending on the location and extent of obstruction [1]. Bouveret syndrome is considered a variant of gallstone ileus, which typically occurs when a gallstone passes from the gallbladder into the small intestine. It is an extremely rare condition, accounting for less

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than 1% of all cases of gallstone ileus. It primarily affects older individuals, particularly those over the age of 70. It is more commonly seen in women than men [2]. Studying and understanding Bouveret syndrome is crucial for healthcare professionals as it can present with atypical symptoms and be easily misdiagnosed. Early recognition of this condition is essential for appropriate treatment, which is challenging and often involves endoscopic retrieval or surgical intervention.

Case presentation

This case report involves a male patient, aged 35, who exhibited symptoms of intense abdominal discomfort, nausea, and vomiting over a 24-h period. The individual experienced identical symptoms of lesser intensity on three separate occasions throughout the preceding 4-month period, during which they resorted to analgesic medication for temporary relief. Upon thorough examination, it was observed that the individual displayed discomfort upon palpation of the abdomen but did not exhibit any signs of rebound tenderness or fluid thrill. Inflammatory alterations were observed in the comprehensive analysis of blood components, while the remaining blood tests yielded results within the established normal parameters. A contrast-enhanced computed tomography (CT) scan of the abdomen was performed, which identified the presence of the Rigler triad, characterized by pneumobilia, a cholecystoantral fistula, and an ectopic gallstone lodged in the gastric antrum. This condition resulted in an obstruction, as depicted in Figs. 1 and 2. The patient's diagnosis of Bouveret syndrome was determined based on the imaging findings.

An upper endoscopy was conducted, revealing the presence of a sizable gallstone located in the gastric outlet. The endoscopic removal of the gallstone was not feasible. During the surgical examination, a substantial stone was found in the gastric outlet that exhibited immobility both proximally and distally. A surgical procedure known as cholecystectomy was conducted, which confirmed the existence of the sizable cholecystoduodenal fistula that was previously detected in the computed tomography (CT) scan. The surgical procedures that were conducted included a partial duodenectomy, a gastrojejunostomy, a cholecystectomy, and a common bile duct exploration with the removal of common bile duct stones. After examining the bile duct, a t-tube was inserted into the bile duct. A subsequent t-tube cholangiogram was conducted five weeks post-surgery, which indicated the absence of strictures or any new stones. The t-tube was extracted 6 weeks postoperatively. Upon doing a check-up 5 months post-operation, it was determined that the patient exhibited a state of good health.

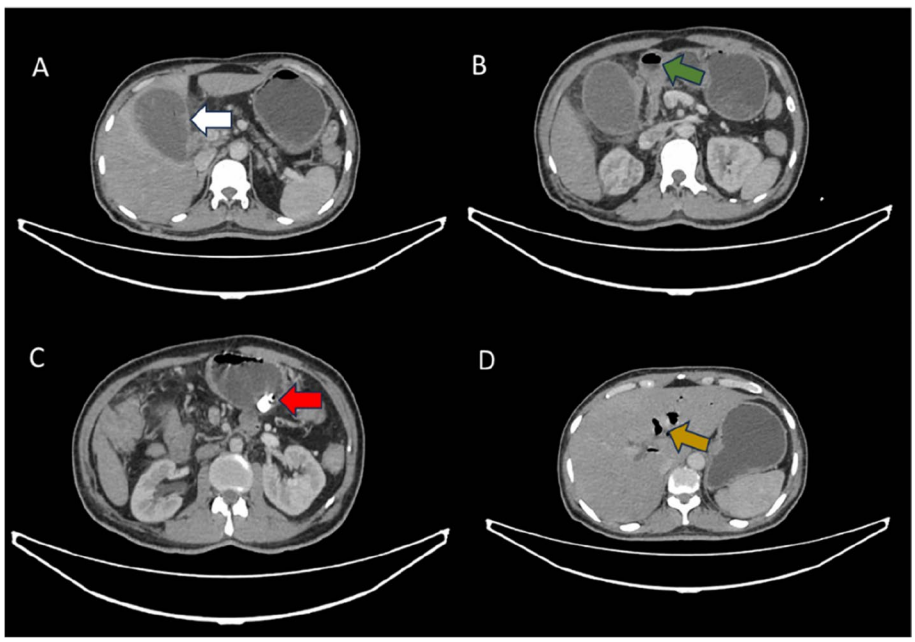

Fig. 1 Axial CECT abdomen: findings suggest Bouveret's syndrome: **A** The gallbladder is distended and inflamed, contains gas, and is in intimate contact with adjacent pylorus and duodenum with loco regional inflammatory changes (white arrow). **B** Cholecysto-antral fistula with ectopic gall stones in the stomach causing gastric outlet obstruction (green arrow). **C** Gastric distension is identified in the pyloric region. Stomach containing calcified gallstones (red arrow). **D** Pneumobilia without biliary tree dilatation (yellow arrow)

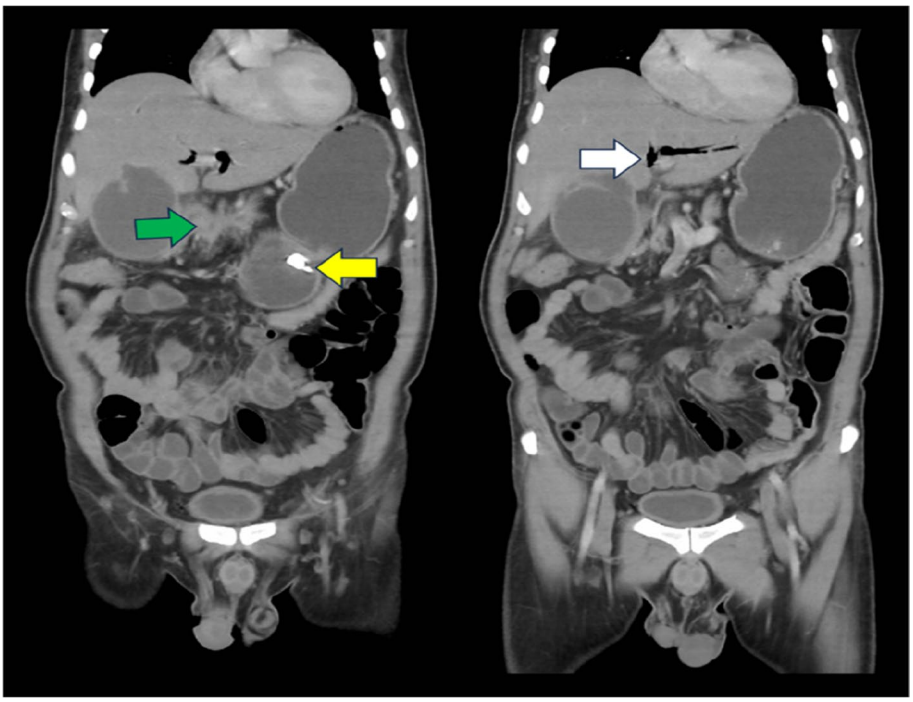

Fig. 2 Coronal view of CT abdominal showing Rigler triad (white arrow indicates pneumobilia. The yellow arrow indicates an ectopic gallstone in the stomach. Green arrow indicates cholecysto-antral fistula causing gastric outlet obstruction)

Discussion

The usual age of presentation for Bouveret syndrome is over the age of 70 and is more commonly seen in women [2], but in this rare case, it is presented in a relatively young male patient with no associated risk factors or comorbidities. Common symptoms experienced by patients with Bouveret syndrome include severe abdominal pain, nausea, vomiting, and bloating. These symptoms can often be mistaken for other gastrointestinal conditions, making an accurate diagnosis challenging [3]. Like in the above case, the patient had frequent less severe attacks of abdominal pain associated with nausea and vomiting, but due to lack of awareness among the medical practitioner, he was diagnosed as a case of food poisoning and was treated with analgesics that subsided the symptoms for the time being. Therefore, healthcare professionals must carefully evaluate the patient's medical history, perform a thorough physical examination, and utilize imaging techniques such as CT scans or endoscopy to confirm the presence of Bouveret syndrome. CT is usually the most practical—its sensitivity for Rigler's triad is between 75 and 78% [4].

Gallstone migration in Bouveret syndrome occurs when a large gallstone from the gallbladder travels through the biliary tract and becomes lodged in the stomach. This obstruction can cause severe symptoms. The impact on the gastrointestinal tract can lead to inflammation, infection, and even perforation if not promptly addressed. Discussion of the specific location of the obstruction and its consequences is crucial in understanding the potential complications of Bouveret syndrome. Depending on where the gallstone becomes lodged, it can lead to further complications such as bile duct obstruction, pancreatitis, or even liver damage [5]. Although rare, it represents a syndrome with an alarmingly high mortality rate, which is estimated between 12 and 30% [6]. Therefore, a thorough evaluation of the location and extent of the obstruction is necessary to determine the most appropriate treatment approach for patients with Bouveret syndrome.

Bouveret syndrome can be treated through surgical and minimally invasive methods. Surgical treatment involves gastric or enteric lithotomy, with or without cholecystoduodenal or choledochoduodenal fistula repair and cholecystectomy. It has higher success rates than minimally invasive options, up to 90% for gastric or enteric lithotomy alone and up to 82% if including fistula repair and cholecystectomy [7]. The decision between single or two operations depends on patient frailty, clinical illness severity, and surgical complexity. According to the literature, a stepwise surgical approach is suggested in older age groups as it reduces the risk of complication, while in the younger age group below 50 years of age, multiple procedures can be performed together depending on

patients' conditions and history [8, 9]. In our case, as the patient had no significant past history or comorbids, so a partial duodenectomy, a gastrojejunostomy, and a cholecystectomy, with common bile duct exploration with the removal of several common bile duct stones along with t-tube insertion, were done in a single surgery and yielded a better healing, prognosis, and minimal complications, hence supporting the aforementioned studies.

Conclusion

Bouveret syndrome is an uncommon cause of gastric outlet obstruction with symptoms similar to various gastric diseases. This illness can be fatal if not diagnosed quickly. A disease usually found in older women was seen in a young boy in the case above. Treatment guidelines for this entity are lacking. The patient's condition, CT scan results, medical history, co-existing illnesses, and intraoperative circumstances determine the next steps. These factors are considered when choosing a single-step or multi-step surgical technique to reduce patient problems. In this example, favorable conditions led to a single-step method with promising results.

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Authors' contributions
MH conceptualized the study and helped in the preparation of the manuscript. MFHQ and FS conducted the literature search and formulated the initial draft of the manuscript. All the authors approved the final version of the manuscript.

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Availability of data and materials
All the data used in this manuscript is included in this paper. If any more information is needed it can be obtained from the corresponding author on reasonable request.

Declarations

Ethics approval and consent to participate
This study is a case report and does not require ethical approval. This case was written in accordance with the Declaration of Helsinki and proper written consent was taken from the patient's parents. No identifying image of information was used in the preparation of this manuscript.

Competing interests
The authors declare that they have no competing interests.

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