International Surgery Journal
Ranjan R et al. Int Surg J. 2023 Apr;10(4):750-753
http://www.ijsurgery.com
pISSN 2349-3305 | eISSN 2349-2902

DOI: https://dx.doi.org/10.18203/2349-2902.isj20230989

Case Report

Rare presentation of gastric outlet obstruction with jejunal impaction of gall stones-the Bouveret's syndrome

Ravi Ranjan*, Gyan Saurabh, Ranvir Singh

Department of Surgery, Lady Hardinge Medical College, New Delhi, Delhi, India

Received: 28 January 2023
Accepted: 02 March 2023

***Correspondence:**
Dr. Ravi Ranjan,
E-mail: ravimaster56@gmail.com

ABSTRACT

Gall stones related complications are very common in the world, Bouveret's syndrome is one of the rare complications (1-3%). Bouveret's syndrome presents a diagnostic and therapeutic challenge. We present a rare case of an 80-year-old lady diagnosed with gastric outlet obstruction with jejunal impaction of gallstones, because of a gallstone impacted in the jejunum 50 cm from duodenojejunal flexure. The patient was operated on with enterotomy and retrieval of gall stones impacted in the jejunum, with roux en y gastrojejunostomy drainage procedure, the gastro-duodenal fistula was left in situ, because of high risk and unstable general condition of our patient intra-operatively. The patient had multiple pre-existing pre and peri-operative comorbidities, fortunately, our patient recovered and our patient was discharged on 21st postoperative days. Identifying Rigler's triad symptoms is synonymous with diagnosing Bouveret's syndrome. However, classical Rigler's triad is present in only 40% of cases, hence alertness and a high degree of suspicion are required to differentiate Bouveret's syndrome from other mechanical causes of gastric outlet obstruction. The atypical cases of Bouveret's syndrome present a challenge to the surgeon for early diagnosis and therapeutic surgical management.

Keywords: Gastric outlet obstruction, Bouveret's syndrome, Rigler's triad

INTRODUCTION

Gallstone is one of the most prevailing surgical diseases in the world with a wide range of clinical manifestations, patients could present from asymptomatic to severe symptoms with systemic complications.[1] Gallstones can present as recurrent biliary colicky pain, acute/chronic cholecystitis, obstructive jaundice with choledocholithiasis with or without severe sepsis and cholangitis, acute/recurrent gallstone induced pancreatitis, Mirizzi's syndrome, gallstone ileus, and gallbladder malignancies.[2] Among them, recurrent biliary pain/acute cholecystitis are the most common complications while gallstone ileus is a rare complication with an incidence of 0.3-0.5% and 1-4% of the causes of dynamic intestinal obstruction.[3] Bouveret's syndrome is a rare cause of gastric outlet obstruction caused due to gallstone complications with impaction of multiple stones/large gall stones in the duodenal bulb after traversing through a billion-duodenal fistula. Causing gastric outlet obstruction is a rare event of a relatively rare disease (gallstone ileus) with an overall incidence of 1-3%.[4] Leon Bouveret, (1896 France) reported two cases of gastric outlet obstruction due to gallstones. Since then, not many cases of Bouveret's syndrome have been reported in the medical literature.[5] If a surgeon is presented with a case of Bouveret's syndrome he/she faces a dilemma for the diagnostic and therapeutic challenges due to its uncommon and unpredictable signs and symptoms.[6]

This case report aims to present a case of multiple gallstones lodging in the mid jejunum, causing mechanical jejunum obstruction presenting as gastric outlet obstruction.

CASE REPORT

An 80-year-old lady presented with complaints of pain upper abdomen for the last 10 days and associated nausea and multiple episodes of vomiting. She had known comorbidities of diabetes mellitus type 2 and hypertension, however, the patient had non-compliance to treatment. On the day of admission, the patient had complaints of upper abdominal pain, nausea and multiple vomiting episodes, which were initially non-bilious followed by several bilious bouts of vomiting, however, the patient had no complaints of fever/loose stools and abdominal examination was grossly normal. The patient was dehydrated, Heart rate was 112 bpm and blood pressure was 110/58 mmHg. She was resuscitated in the emergency ward with iv fluids and foley's urinary catheterized and nasogastric tube insertion, routine blood investigations were sent, and 800 ml of gastric contents were drained by the nasogastric tube. An ABG was done which suggested a picture of metabolic alkalosis (Ph 7.535), and routine blood count demonstrated, anaemia (Hb-7.7) increase of white blood cells (17 K/µl). The serum potassium concentration was 3.2 mmol/l and the serum sodium concentration of 132 mmol/l, with severe hypo-proteinemia (albumin 2 mg/dl) and hypo-calcemia, serum calcium was 6.7 mg/dl. Other blood results of liver function tests and renal functions were normal.

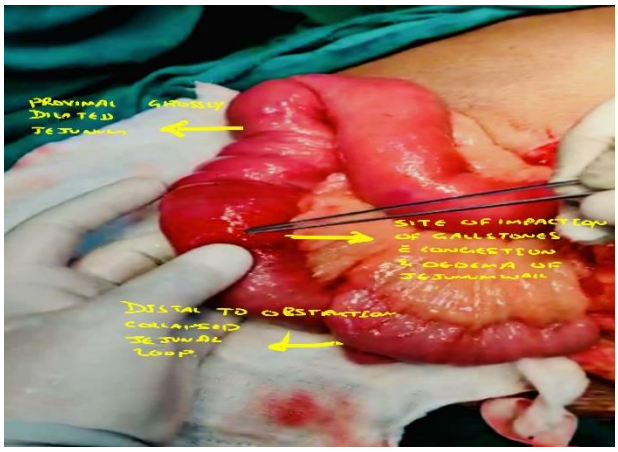

Figure 1: Proximal jejunal gross dilatation with the site of gallstone impaction with distal collapsed jejunum.

With a provisional diagnosis of gastric outlet obstruction, fluid and electrolyte correction was done, and she was initially evaluated with an abdominal ultrasound which suggested chronic cholecystitis with gall bladder wall thickening, the air in biliary radicles, multiple collaterals at porta and a grossly overdistended stomach. Abdominal contrast enhanced computed tomography (CECT) abdomen was planned, which showed contracted gallbladder, fistulous communication between gallbladder and adjacent lateral wall of duodenum first part, air foci in the gallbladder fundus, surrounding omentum showing evidence of fat stranding, air foci within intrahepatic biliary radicles, also CECT abdomen suggested, concentric diffuse oedematous thickening of the distal pylorus and duodenum first part (maximum thickness 13mm), bilateral pleural effusion was noted left side > right side. Upper GI endoscopy was advised, given the advanced age of the patient and pyloric-duodenal wall thickening on CECT Abdomen to rule out malignancy features. Upper GI endoscopy suggested patent pylori duodenal region and a biopsy revealed- inflammatory changes in the pylorus. However, no stones were visualized on either ultrasound abdomen/CECT abdomen/upper GI endoscopy in the duodenum, hence Rigler's triad diagnosis could not be made.

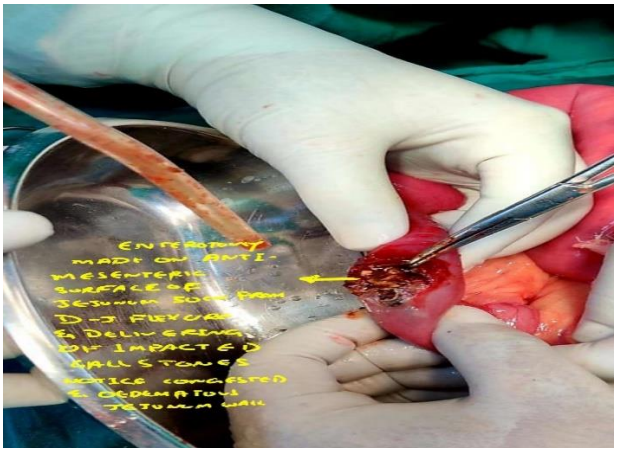

Figure 2: An enterotomy is done on the anti-mesenteric surface of the jejunum with delivering of impacted gall stones.

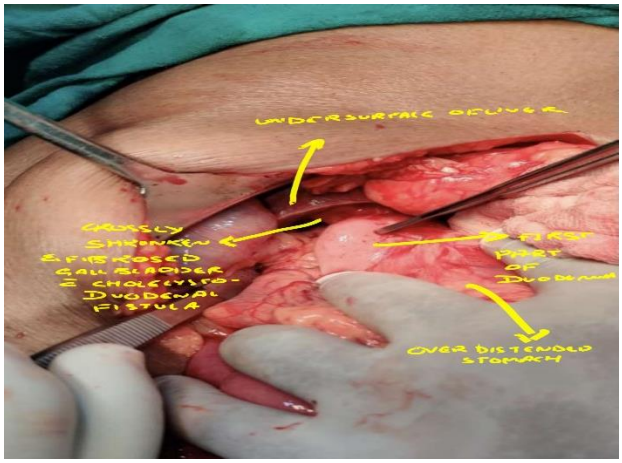

Figure 3: Abnormal cholecysto-duodenal fistula between a grossly fibrosed and shrunken gall bladder and the first part of duodenum with a grossly dilated stomach and a part of liver under surface visible.

The patient was planned for exploratory laparotomy; however, she was unfit to undergo elective surgery, patient 2D ECHO suggested, septal hyperplasia with aortic root dilatation with mild aortic regurgitation with grade 1 diastolic dysfunction, the patient was treated with iv antibiotics for her lung infection (small aspirational pneumonia) and a left side chest tube placed to drain

pleural effusion, and total parenteral nutrition was given to maintain nutrition and correct hypoproteinaemia.

However, with further deteriorating patient condition (ASA IV), with high-risk consent for exploratory laparotomy from the patient's relatives, the patient was operated on in an emergency under general anaesthesia.

Intra-operatively, moderate ascites were found, which were aspirated, gall bladder grossly shrunken and fibrosed, and adhered densely with duodenum first part with a cholecysto-duodenal fistula, stomach and proximal small intestines were grossly dilated, about 50 cm distal to the duodeno-jejunal flexure, a hard mass was palpated, with surrounding small bowel oedematous and unhealthy distal to which small bowel were collapsed, suspecting malignancy an enterotomy was made to look for malignancy and possible frozen section biopsy, how-ever on enterotomy, out-tumbled multiple stones, which had obstructed the jejunum 50 cm from DJ flexure, since the site of obstructed bowel was unhealthy, instead of a simple enterotomy, the affected part of bowel was resected and a roux en y gastro-jejunostomy was made with the firing of staplers, the nasogastric tube was advanced through the jejuno-jejunal anastomosis. Two 28 French drains were placed, one in the right subhepatic region and the other in the pelvis. The fibrosed and shrunken gall bladder with cholecysto-duodenal fistula was left untouched to avoid further morbidity of surgery and unstable ectopic cardiac beats during anaesthesia. The abdomen was closed and the patient was shifted to the intensive care ward mechanically ventilated to assist breathing and correct her physiology, monitor her cardiac condition, post-operative higher antibiotics were continued and total parenteral nutrition was given.

The patient was extubated on postoperative day 3 and shifted to a high-dependency ward on day 5.

Nasogastric tube out-put was minimal on day 5, the oral clear liquid was allowed, and the diet gradually progressed, patient passed stools on day 6 morning. The abdominal drains were removed on postoperative day 12, during the patient's course of hospital stay physician's advice was sought on multiple occasions to correct her chest condition, cardiac condition and blood sugar and hypertension management. The patient was discharged on postoperative day 21, after stabilization of her general condition. The patient's condition was un-eventful for 4 months post-operative period follow-up, however, the patient expired due to sudden heart failure/old age complication in the initial 5th-month post-operative period.

DISCUSSION

Gallbladder stones are a common clinical condition occurring in the general population, women more than men, while the majority of patients are asymptomatic, a small percentage of them develop gall stones related complications, and among them, a few develop serious complications (approximately 6%).[7] Cholecysto duodenal fistula is one such rare gallstone (incidence 1-3%) related complication with fistula formation occurring between the gall bladder and either stomach or duodenum resulting due to chronic inflammation and dense adhesions resulting in decreased arterial blood supply and congestion of venous drainage.[8] As a result, with gradually increasing pressure and necrosis of the chole cysto-duodenal/ gastric wall, gall stones erode into the enteric lumen, which can subsequently migrate proximally or distally, the resulting gastric outlet obstruction is known as Bouveret's syndrome, on radiological imaging has a triad of, the air in biliary radicles, gall stones and small bowel ileus known as Rigler's triad, however, classical Rigler's triad is present in only 40% of patients only.[2] The patients present with clinical complaints of pain abdomen, vomiting multiple episodes with or without features of intestinal obstruction, due to various ages of presentation, a patient may be symptomatic/asymptomatic with a previous history of biliary colicky abdominal pain, in advanced age it may also mimic symptoms of distal gastric malignancy, the patients may present with aspirational pneumonia and other age-related comorbidities.[9] On patient's presentation in emergency features of dehydration and dys-elecrolytemia and Ph must be corrected. Imaging modalities such as CECT abdomen assist in making an early diagnosis of Bouveret's syndrome, upper GI endoscopy must be done before the patient is taken to the operation theatre table to rule out any neoplastic associated/peptic ulcer stricture causes for gastric outlet obstruction.[10] However, surgeons must always have a suspicion of Bouveret's syndrome, whenever a patient with gastric outlet obstruction patient presents with such complaints. On the operation table, the most widely used method is an enterotomy with delivering of the stones from the duodenum/jejunum/ileum and primary repair of the enterotomy. However, the surgeon operating the case is the best judge to assess the bowel whether the bowel is strangulated/congested/grossly oedematous situation and might proceed with the alternate procedure of a simple resection anastomosis or a drainage procedure with roux en y gastrojejunostomy.[11]

CONCLUSION

Bouveret's syndrome is a rare presentation of gall stone induced complications (1-3%). Identification of Rigler's triad symptoms is synonymous with diagnosing Bouveret's syndrome. However classical Rigler's triad is present in only 40% of cases, hence alertness and a high degree of suspicion are required to differentiate Bouveret's syndrome from other mechanical causes of gastric outlet obstruction.[12] The atypical cases of Bouveret's syndrome present a challenge to a surgeon for early diagnosis and therapeutic surgical management.[6]

However, there are no particular established surgical golden rules to predict the migration of gall stones and predict the outcome and severity of the patient's Bouveret syndrome.[11] Most commonly an entero-lithotomy is made

at the site of gall stones impaction and stones are delivered and repaired primarily. In atypical cases, the surgeon on the table involved in the surgery is the best judge to proceed with the type of therapeutic surgery.[13] In our particular case report due to a lack of visualization of stones on various radiological imaging investigations, Bouveret's syndrome diagnosis was not made, and the patient's treatment was delayed to rule out malignancy given the patient's advanced age. Early detection of Bouveret's syndrome could have led to earlier laparotomy and possibly a simple entero-lithotomy would have sufficed, due to the unhealthy nature of the small bowel wall, a roux en y gastro-jejunostomy was made and a multiple unstable ectopic cardiac beat during general anaesthesia led to aborting of resection of cholecystectomy and resection of cholecysto-duodenal fistula with the primary principle of surgery -do no further harm.[14]

ACKNOWLEDGEMENTS

Authors would like to acknowledge the guidance of thesis guide Dr. Manoj Andley, Director professor, Department of surgery LHMC, Delhi; juniors and colleague Dr. Ashutosh and Dr. Shiva who assisted during this high-risk surgical procedure.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

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Cite this article as: Ranjan R, Saurabh G, Singh R. Rare presentation of gastric outlet obstruction with jejunal impaction of gall stones-the Bouveret's syndrome. Int Surg J 2023;10:750-3.