

Bouveret syndrome: Primary demonstration of cholecystoduodenal fistula on MR and MRCP study

Rajkumar Singh Negi, Mukesh Chandra, Rajiv Kapur

Department of Radiodiagnosis, Base Hospital, Delhi Cantonment, New Delhi, India

Correspondence: Dr. Rajkumar Singh Negi, Department of Radiodiagnosis, Base Hospital, Delhi Cantonment - 110 010, New Delhi, India.
E-mail: rajnegi99@hotmail.com

Abstract

Bouveret syndrome is an unusual complication of cholelithiasis which results in upper gastrointestinal obstruction due to a gallstone impacted in the duodenum through a bilio-enteric fistula. We present this rare entity which was primarily diagnosed on magnetic resonance (MR) and MR cholangiopancreatography (MRCP) study.

Key words: Bouveret syndrome; cholecystoduodenal fistula; magnetic resonance cholangiopancreatography; magnetic resonance imaging; MRCP; MRI

Introduction

Bouveret syndrome is a rare form of gallstone ileus which occurs due to impaction of the stone in the duodenum. The usual cause is an underlying bilio-enteric fistula between the gallbladder and duodenum (cholecystoduodenal fistula) through which the stone migrates into the bowel.^[1]

Computed tomography (CT) scan has been useful as a diagnostic modality for demonstration of the obstructing calculus and the bilio-enteric fistula.^[2] We present a case of Bouveret syndrome where primary diagnosis of the cholecystoduodenal fistula along with demonstration of the incriminating calculus was done by magnetic resonance (MR) and MR cholangiopancreatography (MRCP) study of the abdomen.

Case Report

A 72-year-old male presented to the casualty department with complaints of recurrent bilious vomiting and right upper quadrant pain since 15 days. He also gave history of unquantified weight loss since last 3 months. There was no fever, hematemesis, melaena, or jaundice. On physical examination, the patient was frail [body mass index (BMI) 17 kg/m²] and had mild pallor and dehydration. The individual was otherwise anicteric and his vital parameters were stable. Abdominal examination revealed a 6 × 4 cm sized lump in the right upper quadrant which was well defined, smooth, and mobile. His routine blood and urine investigations were within normal limits.

An urgent USG (GE Logiq P5, Color Doppler machine, Korea) of the abdomen was performed which revealed a large calculus possibly impacted in the gut. It appeared as a large hyperechoic lesion casting distal acoustic shadow and was causing proximal bowel dilatation involving the duodenum and stomach. There was evidence of pneumobilia and the gallbladder was not discretely visualized.

CT study of the abdomen was not possible since the patient was vomiting and could not tolerate oral contrast. Therefore, further evaluation of the case was done by MRCP (GE HDx Signa 1.5T MRI System, China). It revealed a 55 × 40 × 38 mm sized hypointense calculus impacted in

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the third part of duodenum with proximal dilatation of the gut [Figures 1 and 2]. The gallbladder was collapsed and revealed thickened walls. A fistulous communication was noted between it and second part of the duodenum on coronal and axial T2-weighted (T2W) images [Figures 3 and 4].

On MRCP, type C biliary variant anatomy with a short right hepatic duct was seen. There was dilatation of the common bile duct (CBD) and common hepatic duct (CHD) seen with evidence of pneumobilia. The cholecystoduodenal fistula was well demonstrated on thick slab images [Figure 5].

Upper gastrointestinal (GI) endoscopy of the patient revealed a cholecystoduodenal fistula in the second part of duodenum. A 5-cm sized calculus was noted in the third part of duodenum beyond which the scope was not negotiable. Repeated attempts at endoscopic retrieval along with extracorporeal shock wave and mechanical lithotripsy proved fruitless and the patient was taken up for surgery.

Per-operatively, the gallbladder was collapsed with dense supracolic adhesions and a frozen C-loop. Enterolithotomy and extrication of the stone was done and a 6 cm sized dirty brown pigment stone was delivered [Figure 6]. The patient showed smooth recovery up to the 5th post-operative day when he developed sepsis and he succumbed to multi-organ system failure on the 10th day.

Discussion

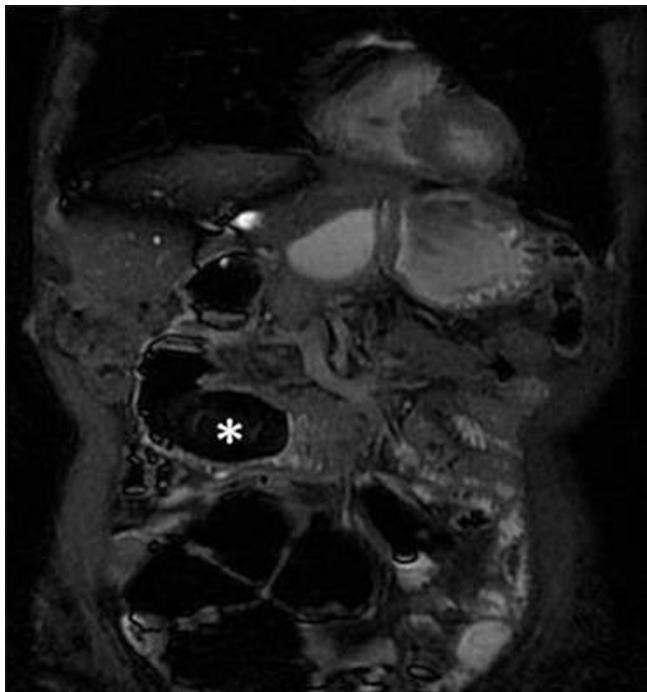


Figure 1: Coronal T2W image showing a large calculus (marked with a star) lodged in the third part of duodenum with proximally dilated gas-filled bowel

Bouveret syndrome was first described by Leon Bouveret in 1896. It is an unusual cause of gastric outlet obstruction due to an impacted gallstone in the duodenum secondary to a cholecystoduodenal fistula. It is a particular form of gallstone ileus which occurs in only 3% of the total cases. Gallstone ileus itself is a rare presentation of the gallstone disease occurring in 0.3-0.5% and constituting 1-3% of all cases of intestinal obstruction.^[1,3]

The rarity of this entity in clinical practice is also evidenced by only up to 300 reported cases in literature.^[4]

Factors favoring the biliary-enteric fistula formation include: size of the gallstone (2-8 cm), long history of biliary disease, repeated episodes of cholecystitis, female sex, and old age (>60 years).^[1] The relative frequency of fistula is cholecystoduodenal (60%), cholecystocolic (15%), cholecystogastric (5%), and choledochoduodenal (5%).^[5]

Once a gallstone erodes into the intestinal lumen, it may manifest as either being asymptomatic, eliminated by either fecal or oral route, or being impacted in the intestinal tract causing obstruction (15% cases).^[6] The obstruction can be as a result of impaction of the stone in the terminal ileum (50-75%), proximal ileum and the jejunum (20-40%), colon and, more rarely, stomach or the duodenum.^[7]

Liew *et al.* observed that the duration of surgical intervention does not influence subsequent prognosis. This is rather influenced greatly by the delay in establishing a correct diagnosis and unjustified delay in intervention. Prompt and correct diagnosis of this entity, therefore, is of paramount importance in managing these patients with a relatively high mortality.^[8]

Historically, abdominal radiograph demonstrating classical

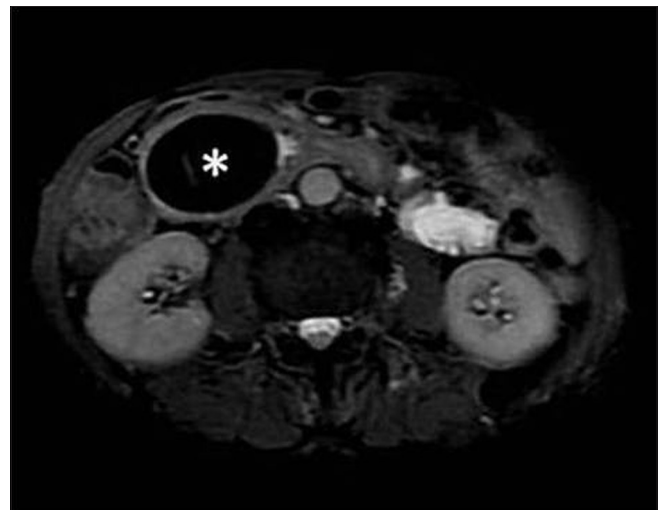


Figure 2: Axial T2W image showing a large hypointense calculus obstructing the third part of duodenum (marked with a star)

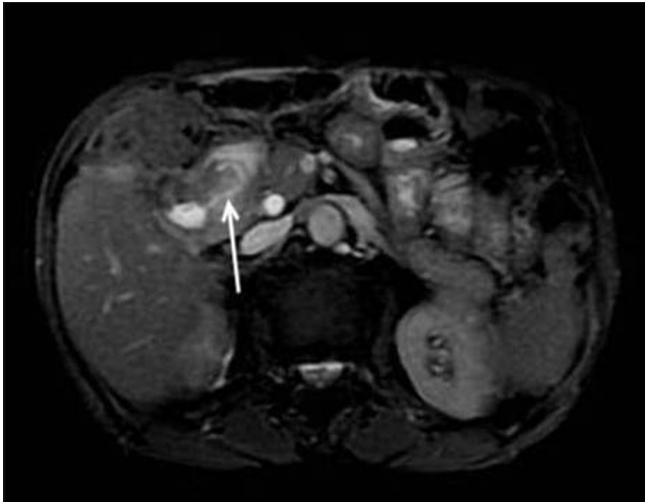


Figure 3: Axial T2W image revealing cholecystoduodenal fistula (marked with arrow). The gallbladder is collapsed, thickened, and adherent to the second part of duodenum

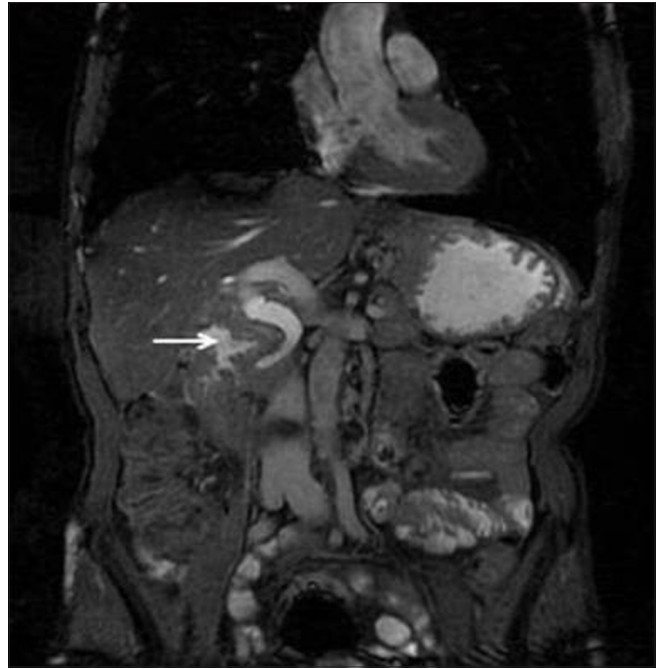


Figure 4: Coronal T2W image revealing cholecystoduodenal fistula (marked with arrow). The gallbladder is collapsed, thickened, and adherent to the second part of duodenum



Figure 5: Thick slab MRCP demonstrating the cholecystoduodenal fistula between the collapsed gallbladder and second part of duodenum. Dilated CBD is also seen

Rigler's triad of bowel obstruction, pneumobilia, and an ectopic gallstone has been described in gallstone ileus. Subsequent radiographs may demonstrate a shift in position of the radio-opaque stone.^[9]

On USG, a dilated stomach with pneumobilia is usually demonstrable. However, the ectopic location of the gallstone within the intestinal lumen may be confused with an orthotopic location in a contracted gallbladder. The fistulous



Figure 6: Per-operative extrication of the incriminating gall stone from the duodenum

tract filled with air or fluid may further be confused with common bile duct.^[10]

CT scan has been useful as an imaging modality for diagnosis of this condition. The Rigler's triad is easily identifiable as is the bilio-enteric fistula which is usually delineated by air or oral contrast. In addition, a secondary sign of presence of contrast in the gallbladder may be a pointer towards the diagnosis.^[9-11]

There is only one reported case by Pickhardt *et al.* so far where a primary diagnosis of the cholecystoduodenal fistula was made on MR and MRCP study of the abdomen. This

may be especially valuable in demonstrating isoattenuating stones and in patients who are unable to tolerate oral contrast medium.^[2]

In our case too, owing to the unstable condition of the patient, a decision to delineate the pathology on magnetic resonance imaging (MRI) study of the abdomen was undertaken with successful demonstration of the impacted gallstone as well as the cholecystoduodenal fistula.

The treatment options include one-stage approach with enterotomy, cholecystectomy, and resection of fistula or a two-stage approach with emergency enterotomy to remove the obstructing gallstone followed by a second stage cholecystectomy after a period of recuperation.^[12] Endoscopic extraction of the gallstone along with extracorporeal shock wave lithotripsy and argon plasma coagulation have also been described as treatment options of this entity.^[13,14]

In our patient, endoscopic extraction of the impacted stone was initially performed failing which the patient was taken up for surgery.

Conclusion

To conclude, Bouveret syndrome is an unusual and a rare complication of gallstone disease. It is associated with a high mortality and the outcome is significantly influenced by timely diagnosis and treatment. MRI and MRCP studies can also act as successful adjuncts in imaging of this disease for primary demonstration of the bilio-enteric fistula in cases where oral contrast is not well tolerated.

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