International Journal of Research in Medical Sciences
Sultana S et al. Int J Res Med Sci. 2023 Jul;11(7):2742-2744
www.msjonline.org
pISSN 2320-6071 | eISSN 2320-6012
DOI: https://dx.doi.org/10.18203/2320-6012.ijrms20232137

Case Report

Atypical presentation of a rare complication of cholelithiasis: Bouveret syndrome

Sadaf Sultana*, Yayati Pimplawar

Department of Radiodiagnosis, Era's Lucknow Medical College and hospital, Uttar Pradesh, India

Received: 25 May 2023
Accepted: 15 June 2023

***Correspondence:**
Dr. Sadaf Sultana,
E-mail: drsadafsultana@gmail.com

ABSTRACT

Bouveret syndrome (BS) is an exceptionally rare variant of gallstone ileus, specified by gastric outlet obstruction resulting from migration of a large gallstone through the bilio-enteric fistula and is impacted in the pylorus or duodenum. We present an atypical case of BS. A 60-year old male came in Era's Lucknow medical college who had undergone multiple diagnostic procedures (x-ray kidney ureter bladder, ultrasonography, computed tomography, magnetic resonance cholangiopancreatography, and endoscopy) (MRCP) for his unusual presentation of right lower abdominal pain without any signs or symptoms of gastric outlet obstruction (nausea or vomiting). Initial attempts of endoscopic retrieval of stone with extracorporeal lithotripsy were performed as first-line treatment which had failed, therefore a successful surgical extraction of stone by enterolithotomy was attempted without cholecystectomy.

Keywords: BS, Gastric outlet obstruction, Gallstone ileus, Endoscopy, Bilioenteric fistula

INTRODUCTION

Bouveret syndrome (BS) is an infrequent complication of cholelithiasis that may cause small bowel obstruction by migrating into the proximal duodenum or pylorus through a bilio-enteric fistula. It was first published by Leon Bouveret in 1896.[1] Risk factors are gallbladder stone (> 2-8 cm), female gender and age >60 years.[2] It is characterized by Rigler's triad (contracted gall bladder, pneumobilia, dilated stomach and ectopic gallstone).[3] The diagnosis is made by endoscopy or imaging. Repairing the fistula is not essential due to its spontaneous closure. Most of the gallstones migrate in the terminal ileum (90%) and few into proximal duodenum (3%).[4]

CASE REPORT

A 60-year-old male presented with the chief complaint of localized pain in right lower abdomen for the past 15 days which was insidious in onset, gradually progressive, non-radiating, and non- aggravating but relieved on taking medication. It was not associated with fever, nausea, vomiting, jaundice, or any co-morbidity. The patient was conscious and oriented. On palpation, his abdomen was soft and non-tender, however, guarding was present in the right lumbar region with no rigidity or rebound tenderness.

His vitals were stable. Blood analysis showed normal white blood cell counts with neutrophilia (75.7%) with normal renal and liver function tests.

The patient was sent to the department of radiodiagnosis to rule out the underlying cause of his pain. X-ray was done as an initial investigation which demonstrated a large staghorn calculus in the left renal fossa irrespective of the site of his complaint (Figure 1).

Subsequently, he underwent contrast-enhanced computed tomography of the abdomen (CECT- KUB) on Siemens Somatom force 384 slice CT scanner to check for any systemic cause, which showed a partially obstructing

staghorn calculus (measuring ~4.7×3.2) cm in the left renal pelvis with completely normal right kidney. We incidentally discovered a contracted gallbladder and a large calculus measuring ~ (2.5×1.9) cm in the duodenal bulb. No communicating fistula was detected to know its cause of emergence (Figure 2).

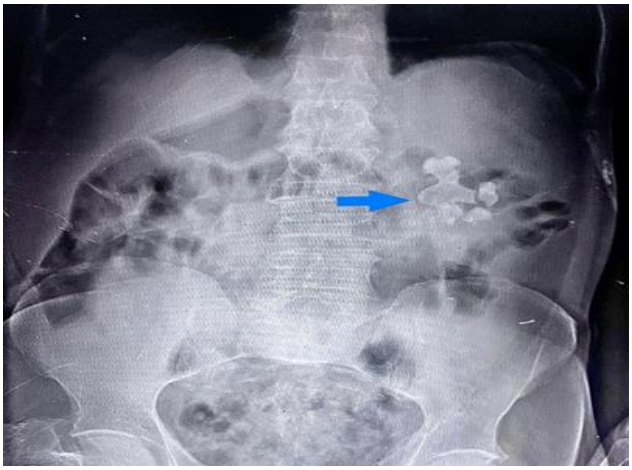

Figure 1: X-ray demonstrates a large staghorn calculus in the left renal fossa (blue arrow).

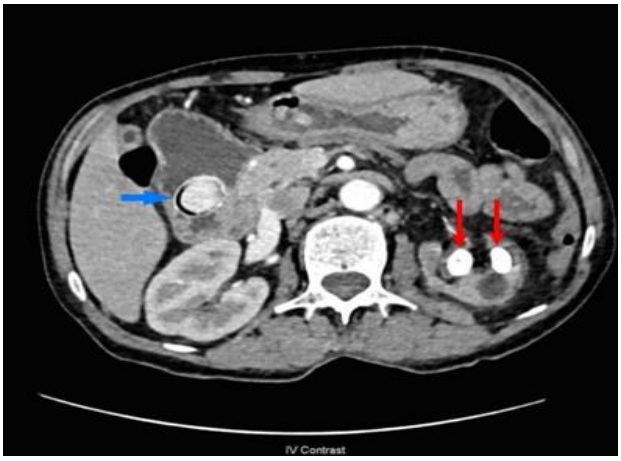

Figure 2: On CECT-KUB (axial view), a staghorn calculus was present in the pelvis extending into the lower calyx of the left kidney (red arrows) with a completely normal right kidney. Contracted gallbladder with a large hyper-dense focus in the duodenal bulb (blue arrow).

Ultrasonography (USG) done on Samsung HS70A machine revealed that the gallbladder was partially contracted and a large echogenic focus casting dense posterior acoustic shadow measuring ~ 2.5×1.9 cm was noted within the proximal duodenum, with no signs of obstruction on the linear array probe (9 MHz). We also found an ill-defined, heterogeneously hypoechoic, loculated collection in the inter bowel space of the right lower abdomen with adjacent mild circumferential bowel thickening with probe tenderness, suspected of infective etiology. This pathology explains the cause of acute pain of the right lower abdomen. Based on the above investigations and findings we suspected the diagnosis of an atypical case of BS without obstruction.

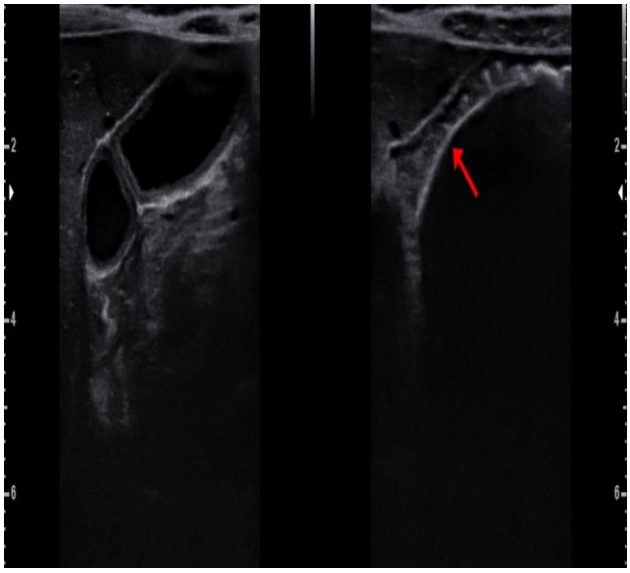

Figure 3: USG revealed a partially contracted gall bladder with a clear lumen on a linear array transducer (9 MHz) and a large echogenic focus casting a dense posterior acoustic shadow in the proximal part of duodenum (red arrow).

Later oral contrast-enhanced MRCP was done on siemens 3 Tesla MRCP which confirmed a large, non-obstructing filling defect in duodenal bulb with no evidence of fistula.

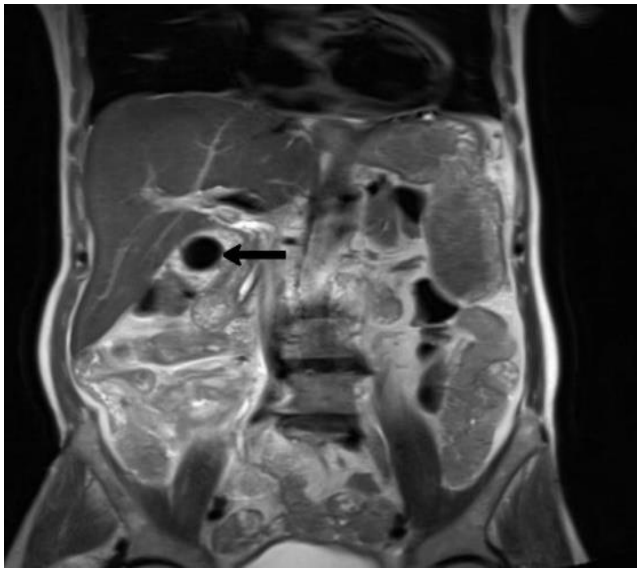

Figure 4: MRCP (coronal view) revealed a large, well-defined, rounded filling defect in the duodenal bulb (black arrow). The defect was not causing gastric outlet obstruction, and there was no evident biliary-enteric fistula found.

After its confirmation, two attempts of (minimally invasive) esophago-gastro-duodenoendoscopy (EGD) were performed using mechanical lithotripsy for stone extraction, but it was irretrievable.

Figure 5: Irretrievable large gallstone in the duodenal bulb on endoscopy.

Therefore, surgical removal of stone was done by breaking it into pieces with the use of Kocher's forceps. An old healed cholecystoduodenal fistula with the gallbladder firmly adherent to the proximal duodenum was also found. Thus, cholecystectomy and fistula repair were not being performed. The patient got discharged on the seventh day after an uneventful recovery without any complications.

As the above findings did not correlate with the chief complaint and the stone was not large enough to cause any obstructive symptoms thus the incidental diagnosis of BS was made.

DISCUSSION

Biliary-enteric fistulas formation occurs by recurrent gall bladder wall necrosis which generally presents as cholecystoduodenal fistula (68%) and rarely as cholecystogastric fistula. It communicates with the duodenum in 85% of cases without causing any gastric outlet obstruction, whereas, can develop an obstruction in 15% of cases.[2]

In the review of literature, it had been stated that although less successful, endoscopy is the most acceptable and least invasive method.[5] Earlier BS was diagnosed based on endoscopy however currently CT is considered the first investigation due to its easy availability.[3] MRCP can be used in patients who are allergic to oral contrast and for iso-attenuating stones on CT, as it differentiates contrast from stone, visualizes fistula, and doesn't require any oral contrast material. Another approach is esophagogastroduodenoscopy (EGD), which has both diagnostic and therapeutic benefits.

CONCLUSION

BS is an uncommon manifestation of gallstone ileus with or without gastric outlet obstruction, where the patient can atypically present with a non-obstructing gallstone if the size is not large enough as in our case. Surgery like gastric or duodenal enterolithotomy with or without bilio-enteric fistula repair and cholecystectomy is the mainstay of treatment, although repairing of a spontaneously healed fistula is not required. Rapid diagnosis and timely removal of stones are important steps for improvement of prognosis and reduction in mortality.

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

REFERENCES

1. Yu YB, Song Y, Xu JB, Qi FZ. Bouveret's syndrome: A rare presentation of gastric outlet obstruction. Exp Ther Med. 2019;17(3):1813-6.
2. Turner AR, Kudaravalli P, Al-Musawi JH, Ahmad H. Bouveret Syndrome (Bilioduodenal Fistula). In: StatPearls. Treasure Island (FL): StatPearls Publishing. 2022;28613489.
3. Brennan GB, Rosenberg RD, Arora S. Bouveret syndrome. Radiographics. 2004;24(4):1171-5.
4. Joshi D, Vosough A, Raymond TM, Fox C, Dhiman A. Bouveret's syndrome as an unusual cause of gastric outlet obstruction: a case report. J Med Case Rep. 2007;1:73.
5. Yang KJ, Chang CK. Bouveret Syndrome: A Rare Case of Instance and Treatment in a Younger Patient. Case Rep Gastrointest Med. 2020;2020:1837387.

Cite this article as: Sultana S, Pimplawar Y. Atypical presentation of a rare complication of cholelithiasis: Bouveret syndrome. Int J Res Med Sci 2023;11:2742-4.