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A new variant of Bouveret's Syndrome

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Abstract

Gallstone ileus is rare and mostly involves the terminal ileum. When it occurs in the duodenum or gastric outlet, it is known as Bouveret's syndrome; which is the rarest form of gallstone ileus. We describe a case report of gastric outlet obstruction with a small solitary ectopic gallstone in the pylorus simulating Bouveret's Syndrome. This is a case of a middle-aged man, presented with abdominal pain, concomitant non-bilious vomiting, and fever. The case report describes the clinical presentation and imaging findings.

Keywords

Bouveret's syndrome; chronic cholecystitis; gastric outlet obstruction; gallstone ileus

Abbreviations

OGDS: Oesophagoduodenoscope; CT: Computed Tomography

Case Presentation

A case of 55-year old male patient presented to emergency department with one-week history of right sided upper abdominal pain with concomitant multiple episodes of non-bilious vomiting and fever. He had several episodes of previous right hypochondria pain for the past one year but did not sought medical help. No alteration of the normal bowel habit. Clinical evaluation noted right hypochondria tenderness with no clinical evidence of jaundice. Blood investigations revealed leukocytosis (WCC 10.4 x 10^9/L). The patient was manages as an acute-on-chronic cholecystitis with intravenous fluids and antibiotics. An abdominal ultrasound revealed a dilated gallbladder with thickened wall, presence of pericholecystic fluid and grossly dilated stomach.

Further evaluation with abdominal contrast enhanced computed tomography (CT) revealed an irregular, dilated gallbladder with thick wall and surrounding streakiness suggestive of inflammatory changes. The stomach is grossly dilated with a small stone (6 mm x 6 mm) embedded in the stomach

pylorus (Figure 1A and 1C). The pylorus itself is thick. The combination of the inflammatory changes of the gallbladder and the pylorus result in gastric outlet obstruction (Figure 1B and 1D). The rest of the small and large bowels were normal. There is no distal bowel obstruction or pneumobilia. Subsequently, the initial oesophagoduodenoscopy (OGDS) evaluation performed three days after the acute admission. It showed reflux esophagitis, but no fistula demonstrable. The patient then managed conservatively with antibiotics and discharged well six days later.

An outpatient CT scan reassessment done 6-weeks later after completion of antibiotic showed, features suggestive of chronic cholecystitis with no residual stone or gastric outlet obstruction seen (Figure 2). Repeated OGDS done 4 months after the initial presentation revealed an ulcer with elevated surrounding mucosa at D1 region. There is also adjacent fistulous opening seen with presence of bile effluent (Figure 3). This represents a cholecysto-duodenal fistula.

Finally, he undergoes an open cholecystectomy 5 months later. Intraoperatively reveal dense adhesion of gallbladder neck to the duodenum. A fistula tract with obstructing gallstone seen, at the neck of gallbladder (Figure 4).

Discussion

Gallstone ileus is rare, representing of 1-4% of all small bowel obstruction [1], which mostly occurs in the terminal ileum. In descending order of frequency, the stone may be seen in the proximal ileum, distal jejunum, colon and the duodenum or gastric outlet; also known as Bouveret's syndrome [2,6,7]. Imaging findings of Rigler's triad is helpful for diagnosis, by having at least 2 out of 3 features though it can only demonstrate up to 77.8% in Bouveret's syndrome with CT [5]. Rigler's triad consists of dilated bowel, ectopic stone, and pneumobilia. Stones associated with Bouveret's syndrome are typically greater than 2.0-2.5 cm in size following the general rule that states: the larger the stone, the more proximal the obstruction [3,4]. This occur as a result from stone that migrates through a cholecystoenteric fistula [8-11] .

In our case, however, we report a proximal gallstone ileus with the offending stone being solitary and lesser than 2.0 cm. To our knowledge from Pubmed search, to date, no such case ever been reported. This will be the first case report of proximal gallstone ileus with the stone smaller than 2.0 cm. For this reason, we report this as a new variant of Bouveret's Syndrome. Stomach dilatation with ectopic gallstone in the pylorus was the two findings of Rigler's triad seen in our case, while missing the pneumobilia.

The possible pathogenesis is, chronic cholecystitis causes a cholecysto-duodenal fistula formation that allows persistant duodenal exposure towards bile and gallstone to pass through. This will then leads to the formation of duodenal ulcer, and inflammation of the surrounding duodenum causing an intraluminal narrowing. In addition, the inflammation of the gallbladder during acute attack further contributes to the narrowing of the pylorus by external compression. Together with the small stone, these finally result in gastric outlet obstruction. As the fistulous track is small, it can easily be miss in the initial OGDS as well as on the CT scan; as what had happened in our case. Thus, we propose that a repeated OGDS is warrant in such cases.

Furthermore, the patient recovered well after conservatively managed with a course of antibiotic. This explained that after the inflammation settled, the stone dislodged and passed out through the feces. An interval CT scan done six-weeks later confirmed no residual of stone in the alimentary system. Cholecystectomy is still the final treatment of choice to prevent recurrence.

Conclusion

Bouveret's Syndrome is rare and is usually associated with large stones. In this new variant of Bouveret's syndrome, we describe proximal gallstone ileus with a small, solitary stone associated with cholecystoduodenal fistula and concomitant gastric outlet obstruction secondary to inflammation and edema of the pylorus and the gallbladder. Therefore, clinician and radiologist should be aware of this variant in patients presented with gastric outlet obstruction.

Figures

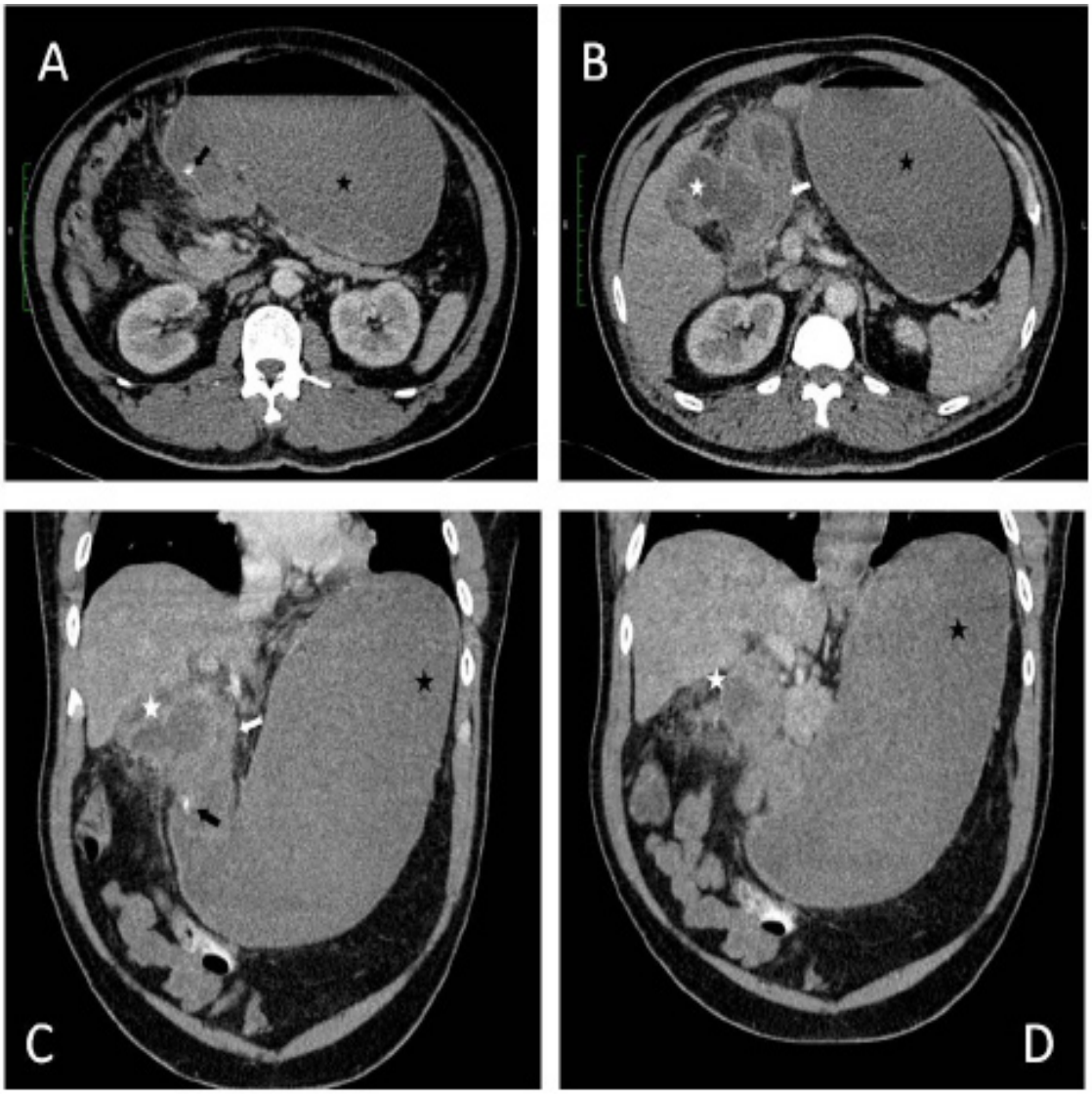

Figure 1: Axial CT abdomen (A and B) and coronal oblique (C and D) showed a dilated stomach (★) with small embedded stone (6x6mm) (black arrow) with inflammatory changes at the pylorus and gallbladder (☆) resulting in gastric outlet obstruction (white arrow).

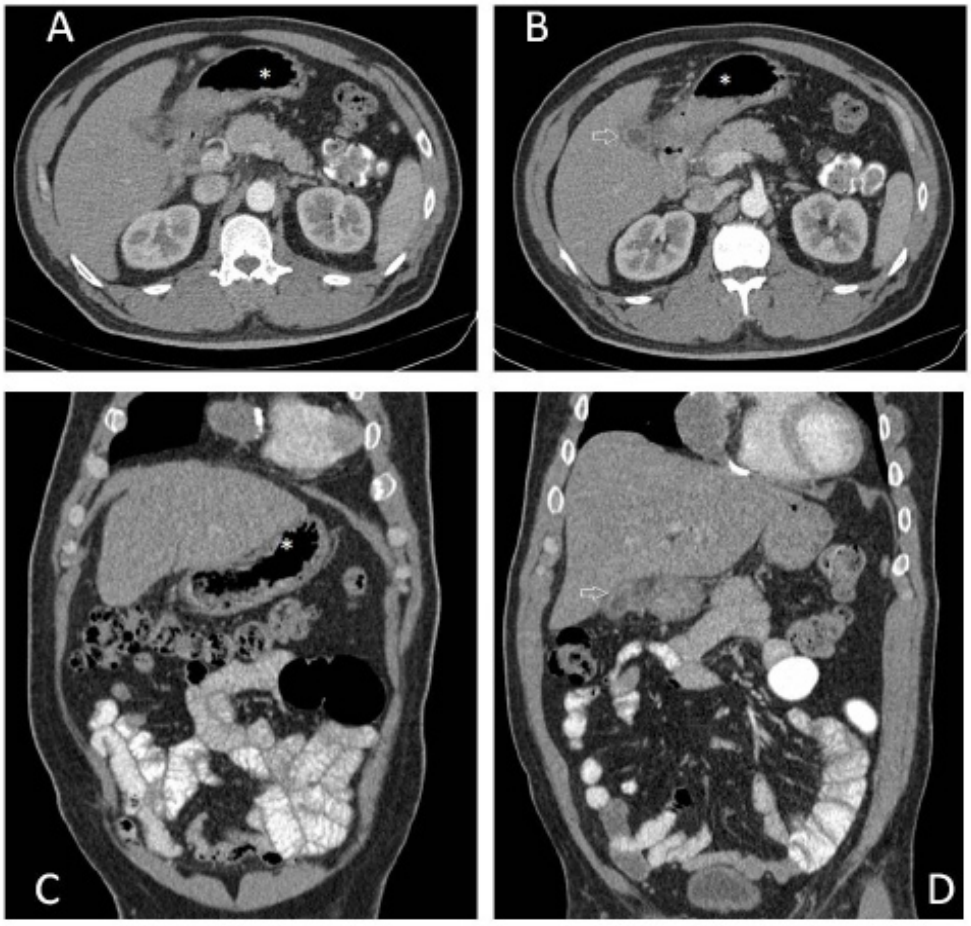

Figure 2: A repeat CT abdomen in axial (A and B) and coronal (C and D) sequences showed non-dilated stomach (*). The gallbladder (arrow) is mildly distended with thickened wall and presence of pericholecystic fluid. No gallstone seen.

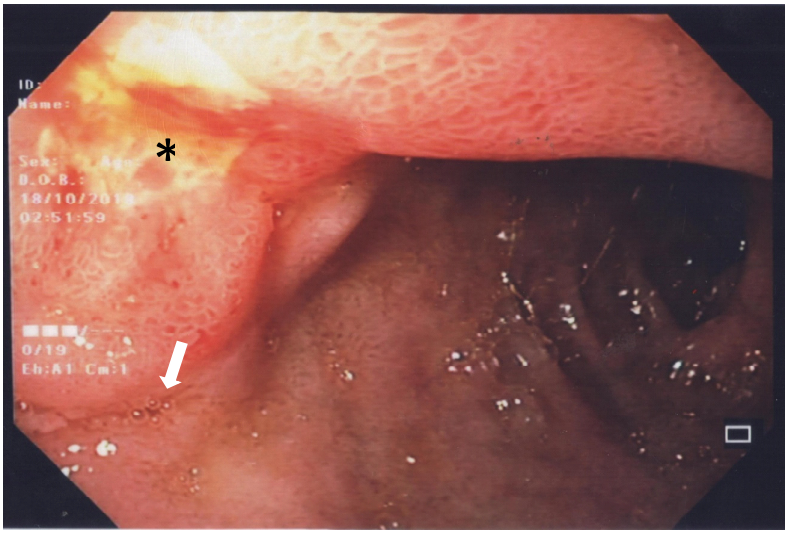

Figure 3: Oesophagoduodenoscope (OGDS) noted D1 ulcer with elevated surrounding mucosa (*) with adjacent opening seen containing bile effluent (white arrow).

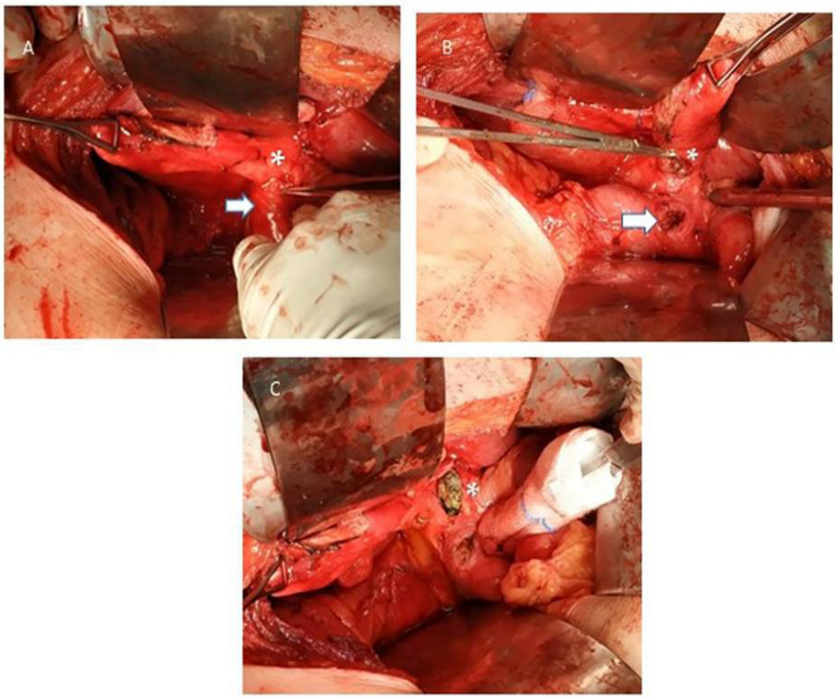

Figure 4: Intraoperative images showing (A) gallbladder neck (*) adherent to duodenum (white arrow). (B) Separation of gallbladder neck (*) from duodenum demonstrated the fistulation tract (white arrow). (C) The gallstone causing fistulation seen after opening the gallbladder at the neck (*)

References

1. Clavien PA, Richon J, Burgan S, Rohner A. Gallstone ileus. British Journal of Surgery 1990;77:737–42.

2. Nuno-Guzman CM, Arroniz-Jauregui J, Moreno-Perez PA, Chavez-Solis EA, Esparza-Arias N, Hernandez-Gonzalez CI. Gallstone ileus: one-stage surgery in a patient with intermittent obstruction. World J Gastrointest Surg. 2010;2(5):172–176.

3. Koulaouzidis A, Moschos J. Bouveret's syndrome. Narrative review. Ann Hepatol. 2007;6(2):89–91.

4. Patel A, Agarwal S. The yellow brick road of Bouveret syndrome. ClinGastroenterolHepatol 2014; 12: A24.

5. Lassandro F, Gagliardi N, Scuderi M, Pinto A, Gatta G, Mazzeo R. Gallstone ileus analysis of radiological findings in 27 patients. Eur J Radiol 2004; 50(1):23–29.

6. Reisner RM, Cohen JR (1994) Gallstone ileus: a review of 1001 reported cases. Am Surg 60: 441-446.

7. Mishra A, Jain A, Lal P, Hadke NS (2013) Bouveret Syndrome: A Case Report and Review. J Gastroint Dig Syst 3:133.

8. Gencosmanoglu R, Inceoglu R, Baysal C, Akansel S, Tozun N. Bouveret's syndrome complicated by a distal gallstone Ileus. World J Gastroenterol 2003; 9(12): 2873-2875.

9. Ajay K. Singh, Ali Shirkhoda, Nirish Lal, and PallaviSagar, Bouveret's Syndrome: Appearance on CT and Upper Gastrointestinal Radiography Before and After Stone Obturation. American Journal of Roentgenology 2003 181:3, 828-830.

10. Ghazi RajiQasaimeh, SohailBakkar, Khaled Jadallah. Bouveret's Syndrome: An Overlooked Diagnosis. A Case Report and Review of Literature. Int Surg. 2014 Nov-Dec; 99(6): 819–823.

11. AL-Habbal Y, Ng M, Bird D, McQuillan T, AL-Khaffaf H. Uncommon presentation of a common disease - Bouveret's syndrome: A case report and systematic literature review. World J GastrointestSurg 2017; 9(1): 25-36.

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