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The Bouveret's Syndrome: A Case Report of a Rolling Gallstone from the Gallbladder to the Duodenum

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Abstract

We report a case of a 92-year-old man complaining of epigastric pain, which an US first and a CT scan later revealed to be related to a large gallstone causing cholecystitis. After the patient had refused surgical treatment for this condition, he was again referred to our Emergency Department presenting with a clinical picture of gastric obstruction. A new CT scan showed the classic Rigler's triad, characterized by pneumobilia, gastric distension and gallstone in the duodenal lumen, which was pathognomonic for a rare form of gallstone ileus named Bouveret's syndrome. The cause of this event was found out to be the chronic inflammation of gallbladder wall and its consequent erosion, which led to formation of gallbladder-duodenum fistula and the movement of the gallstone from the gallbladder to the duodenum where it impacted. This is a high morbidity and mortality condition, which affects mostly elderly people and needs early diagnosis and surgical treatment.

Keywords

Cholecystitis, Cholelithiasis, Gastric Outlet Obstruction, Cholecystoduodenal Fistula, Ileus

1. Introduction

The Bouveret's syndrome is a rare and high-risk complication of cholecystitis, affecting only 0.9% - 3.2% of patients with history of gallstones [1]-[3]. It consists in a gastric outlet obstruction due to the development of an internal biliary fistula and the migration of gallstones from the gallbladder to the bowel [4] [5]. Once the gallstone passes in the duodenum, it may cause a mechanical obstruction at the level of the ileum, or less commonly,

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a gastric obstruction. Correlation of a non-specific clinical picture with imaging findings is important to achieve the correct diagnosis and the right therapeutic approach. Many authors demonstrated that US and plain film was usually inconclusive, whereas CT was an effective technique demonstrating not only the gastric obstruction due to the gallstone, but also the biliary-enteric fistula [5]-[11].

To our knowledge, there are not many cases in the literature describing, as in our case report, the natural history of Bouveret's syndrome, from cholecystitis to biliary fistula and gastric outlet obstruction in the same patient. In addition, main characteristics are presented with clear CT images.

2. Case Report

A 92-year-old male in poor general condition was referred to our Emergency Department presenting epigastric pain. He already suffered from moderate hypertension, renal impairment and diabetes and also complained of sporadic episodes of aspecific abdominal pain.

The abdominal radiography showed an area of increased opacity in the right upper quadrant, measuring about 4 cm (**Figure 1**).

An ultrasound examination (US) was then performed to evaluate the cholecystic area and it demonstrated a curvilinear focus of increased echogenicity that measured approximately 5 cm with posterior acoustic shadowing (**Figure 2**), suggestive of cholelithiasis. However, further examinations were needed as the cholecystic wall was not completely identified. An abdominal contrast-enhanced computed tomography (CECT) revealed the presence of signs of gallstone-related cholecystitis (**Figure 3**).

The patient refused the surgical treatment offered and, after few weeks, he was again admitted to the Emergency Department with signs of intestinal obstruction. The CECT scan performed at this stage showed a gallstone in the duodenal lumen (**Figure 4**), gastric distension and pneumobilia (**Figure 5**). Furthermore, the oral administration of water immediately before the scan allowed identifying a fistula from the gallbladder fossa toward the second part of the duodenum (**Figure 4**). These findings were in keeping with the hypothesis of gastric outlet obstruction due to a large gallstone impacted in the duodenum.

Because of his comorbidities, a surgical approach with enterolithotomy alone was preferred.

Gallstone induced ileus is a rare complication of cholelithiasis and duodenal outlet obstruction is an even more rare variant defined Bouveret's syndrome.

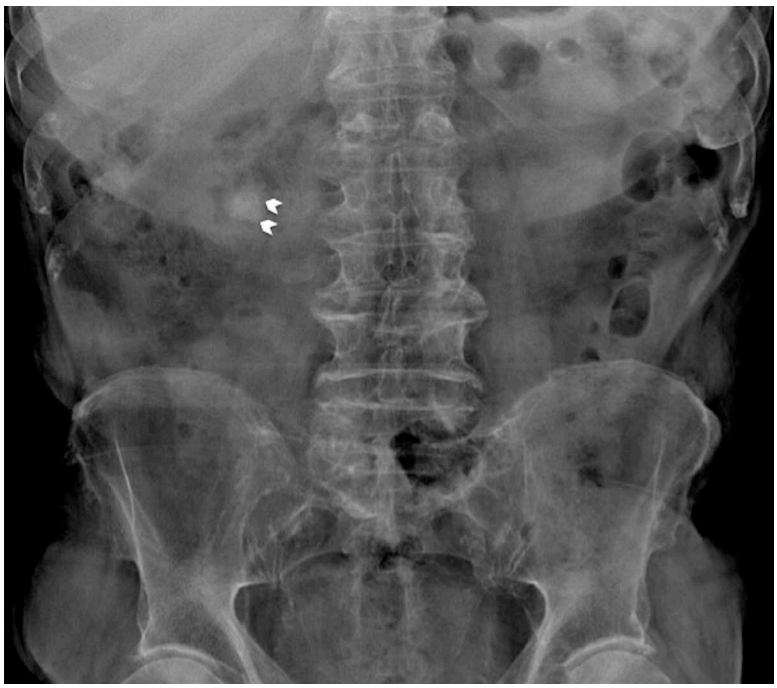

Figure 1. The abdominal radiography shows an area of increased opacity in the right upper quadrant measuring about 4 cm (arrowhead).

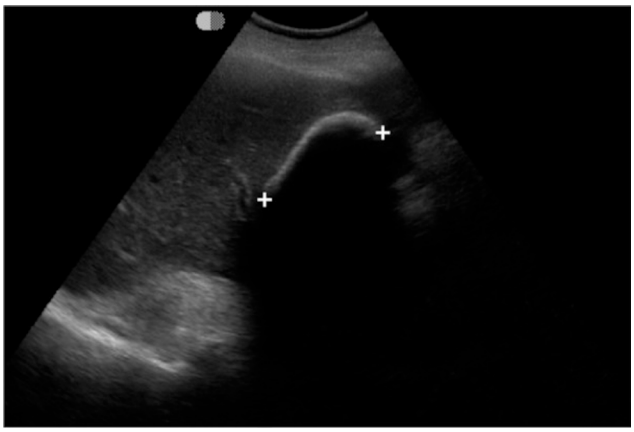

Figure 2. The abdominal ultrasound examination demonstrates a curvilinear focus of increased echogenicity that measures approximately 5 cm with posterior acoustic shadowing.

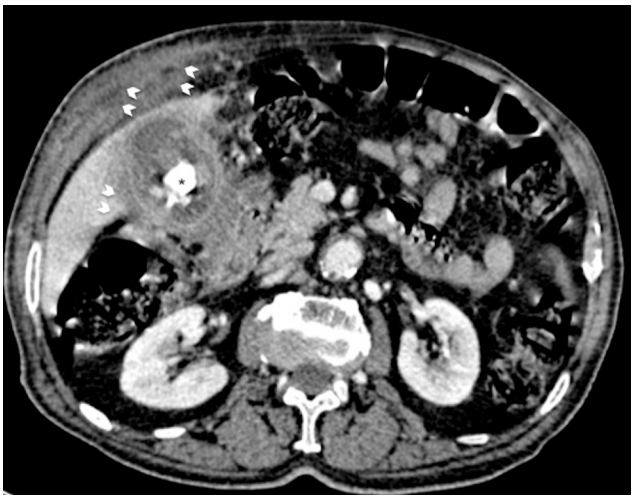

Figure 3. The abdominal contrast-enhanced computed tomography shows the presence of fluid effusion (arrowheads) and a gallstone in the cholecystic lumen (asterisk).

Figure 4. The abdominal contrast-enhanced computed tomography shows a gallstone in the duodenal lumen (asterisk) and the fistula (arrow).

Figure 5. The abdominal contrast-enhanced computed tomography shows gastric distension and pneumobilia (arrowheads).

3. Discussion

Gallstone disease is not uncommon, affecting about 10% of population in the United States and Western Europe. However, only 20% - 30% of patients will experience biliary pain or complications such as acute cholecystitis, cholangitis or pancreatitis [1] [2].

Another possible complication is the development of an internal biliary fistula. This rare condition, affecting 0.9% - 3.2% of patients with biliary disease [3], is due to the close anatomic relationship between the gallbladder and the duodenum and the proximal transverse colon. As a result, chronic inflammation of the gallbladder may lead to chronic perforation and fistulous communication to these structures [4]. In a series of 10 patients Oikarinen *et al*. [5] demonstrated that acute or chronic cholecystitis is a significant predisposing factor of internal biliary fistula and that, as it happened in the case described above, the most frequent is the cholecysto-duodenal type. Once a fistulous tract has established, stones may pass from the gallbladder into the bowel and, if greater than 2 - 2.5 cm, may cause mechanical obstruction, mostly at the level of the ileum, resulting in the so-called gallstone ileus. Less common event is the obstruction at the level of the gastric outlet or duodenum, which represents a specific subset of gallstone ileus and is referred to as Bouveret's syndrome [1] [4]-[7].

Bouveret's syndrome is more prevalent in the elderly and in females, with a reported median age of 74 years and a female to male ratio of 1.9 [1] [2] [5]. An early diagnosis of this condition is recommended, as its mortality rate remains as high as 12%, nevertheless it has declined over recent years [7]-[9]. The clinical picture is non-specific, with signs and symptoms of gastric outlet obstruction [10], therefore, correlation with imaging findings is needed to achieve the correct diagnosis [2] [8] [9].

Imaging of the abdomen by plain x-rays is the appropriate initial step but is, alone, diagnostic of Bouveret's syndrome in only 21% of cases [8]. Pneumobilia seen on imaging studies strongly suggests the presence of an internal biliary fistula in the absence of prior sphincterotomy, surgical bypass procedure, recent endoscopic retrograde cholangiopancreatography, or passed common duct stone. The Rigler's triad of small-bowel obstruction, pneumobilia and ectopic gallstone (s) is virtually pathognomonic for gallstone ileus but is detected on plain films in only 30% - 35% of patients [2] [4]-[8] [10].

This triad of findings, however, is more often apparent on CT, which is an effective imaging technique in this clinical setting [5]-[11]. In addition, as demonstrated by Pickard *et al*. [6], CT can also provide important information on the degree of bowel obstruction and suggest the likely site of fistula. This latter may be seen if the tract is enhanced by positive oral or air contrast material. A secondary sign that may be useful is the identification of oral contrast material within the gallbladder [4] [7]. One potential drawback of CT is that 15% - 25% of gallstones appear as isoattenuating relative to bile or fluid. MRCP may be useful in such situation [10]. In our case, the CT scan we performed, showed a high attenuating gallstone in the duodenum and the prior gastric distension with water led to clearly identify the fistula.

A prompt relief of the obstruction is the main goal to achieve, to limit the consequent fluid and electrolytes impairment. However, surgery often is not desirable treatment as patients are often poor surgical candidates secondary to concomitant illnesses and advanced age. On the contrary, many studies suggest that the main treatment modality for Bouveret's syndrome is surgical approach. In the elderly, enterolithotomy alone may be adequate treatment and subsequent cholecystectomy may not be required as a spontaneous resolution of the fistula is expected at this point [2] [3] [7] [8].

4. Conclusion

Our case demonstrates the development of enterobiliary fistula as a rare complication of cholecystitis. In addition, the significant size of the gallstone led to an even rare condition of gastric obstruction, named Bouveret's syndrome. Because of its high morbidity and mortality, an early diagnosis is important. The classic Rigler's triad could be appreciated on abdominal radiography, or more commonly, on CT scan. As a consequence, in a patient with known recent history of cholecystitis, presenting with clinical picture of intestinal obstruction, a Bouveret's syndrome has to be suspected. A CT scan is mandatory, even better after gastric distension with water, to confirm the presence of a fistula. A surgical approach should be preferred even if there is no agreement among authors on the choice of the best procedure.

Acknowledgements

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