BOUVERET'S SYNDROME – A RARE CAUSE OF INTESTINAL OBSTRUCTION

SÍNDROME DE BOUVERET – UMA CAUSA RARA DE OBSTRUÇÃO INTESTINAL

Urânia FERNANDES[1], Gonçalo GUIDI[1], Daniela MARTINS[1], Rita MARQUES[1], Herculano MOREIRA[1], João PINTO-DE-SOUSA[1]

[1] General Surgery Department, Hospital Center of Trás-Os-Montes and Alto Douro (CHTMAD), Clinical Academic Centre of Trás-os-Montes and Alto Douro (CACTMAD); Portugal

Correspondence: Urânia Fernandes (ufernandes@chtmad.min-saude.pt)

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ABSTRACT

This report is about a clinical case of Bouveret's Syndrome (BS), a gastric outlet obstruction by a gallstone. It is rare and often a delayed diagnosis in elderly women with comorbidities. These factors contribute to a relevant morbidity and mortality, underlying the importance of an adequate and timely treatment. Its inespecific simptoms makes a high clinical suspicion essential. In the presence of Rigler's triad the plain abdominal X-ray is diagnostic in one third of the cases but the diagnosis is generally made by CT-scan or esophagogastrosduodenoscopy. This one is often attempted first but surgery is almost always needed. Enterolithotomy/gastrotomy can be complemented by cholecystectomy in a one or two-stage surgery. The most definitive surgical procedures should be restricted for patients in the best physiological conditions.

Keywords: *gallstone, ileus, intestinal obstruction.*

RESUMO

Este é um caso clínico sobre Síndrome de Bouveret (SB), que consiste na obstrução do antro gástrico por um cálculo biliar. É um diagnóstico raro e frequentemente tardio, principalmente em mulheres idosas com múltiplas comorbilidades. Todos estes fatores contribuem para uma importante morbimortalidade, o que realça a importância de um tratamento adequado e atempado. A inespecificidade da sua apresentação clínica torna essencial uma elevada suspeição clínica. A presença da tríade de Rigler na radiografia abdominal é diagnóstica em cerca de dois terços dos casos, mas geralmente o diagnóstico é obtido após realização de tomografia computorizada abdominal ou endoscopia digestiva alta. Esta última é ocasionalmente terapêutica, mas a cirurgia é geralmente o tratamento definitivo. A enterolitotomia ou gastrotomia podem ser complementadas com colecistectomia simultânea ou diferida. Procedimentos cirúrgicos mais invasivos devem ser reservados para doentes com melhores reservas fisiológicas.

Palavras-chave: *litíase, ileus, oclusão intestinal.*

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INTRODUCTION

The prevalence of gallstone disease varies in Europe (5.9%-21.9%) and acute cholecystitis is the commonest complication.[1,2] Gallstone Ileus (GI) occurs when a gallstone causes intestinal obstruction. It is a rare complication (0,3-0,5%) but in patients older than 65 years it is responsible for 25% of all cases of small bowel obstruction. Bouveret's syndrome accounts for 3% of all cases of GI and occurs when a gallstone impacted in duodenum causes gastric outlet obstruction.[3] Its nonspecific symptoms make a high index of clinical suspicion essential for diagnosis and adequate treatment. It is often a delayed diagnosis, occurring mostly in elderly women with comorbidities. These factors contribute to significant morbidity and mortality.

CASE REPORT

A 63-years-old male presented in the emergency department with a four-day history of epigastric pain, vomit and diarrhea. Past medical history of the patient included schizophrenia, arterial hypertension, hyperlipidemia and chronic gastritis. Two months prior, he had been admitted for a self-limited duodenal obstruction caused by a bezoar. He was now emaciated, dehydrated, icteric and normothermic. His blood pressure was 79/40 mmHg. He had halitosis and referred pain with epigastric palpation but without peritoneal irritation signs.

Laboratory results revealed elevation of inflammatory parameters and acute renal disfunction (table 1).

Plain abdominal X-Ray showed an ectopic gallstone, small bowel distension but no pneumobilia (figure 1). Esophagogastroduodenoscopy (EGD) identified reflux esophagitis, a 5 millimeters clean base ulcer in the lesser curvature and an unremovable foreign body in the bulb. Abdominal CT-scan revealed acute cholecystitis, a gallstone in the second duodenal portion with 3 centimeters (cm)

Table 1 – **Laboratory results.**

Leucocytes (neutrophils)	22700/uL (85,9%)
Creatinine	2,2 mg/dL
BUN	131 mg/dL
RCP	2,65 mg/dL
CPK	393 U/L
No hepatic alterations	

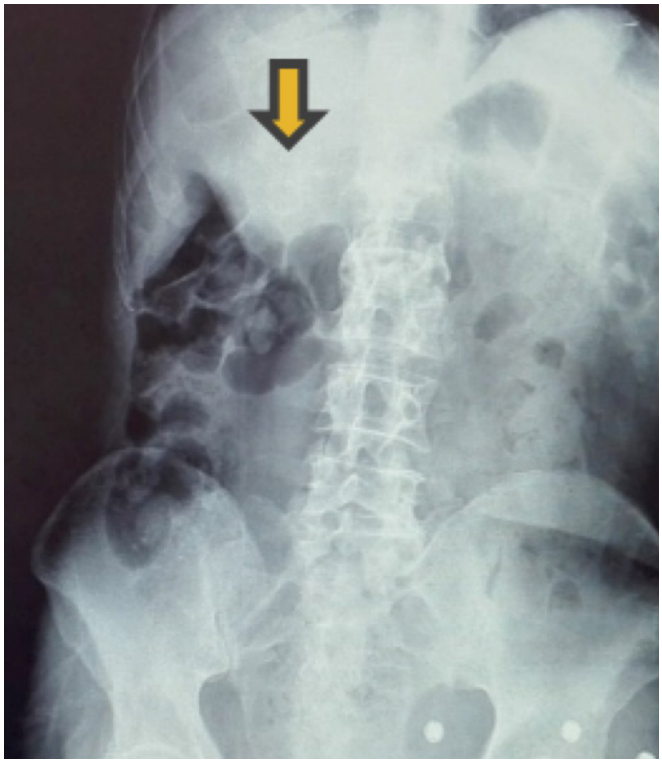

Figure 1 – **Ectopic gallstone (arrow) in X-Ray.**

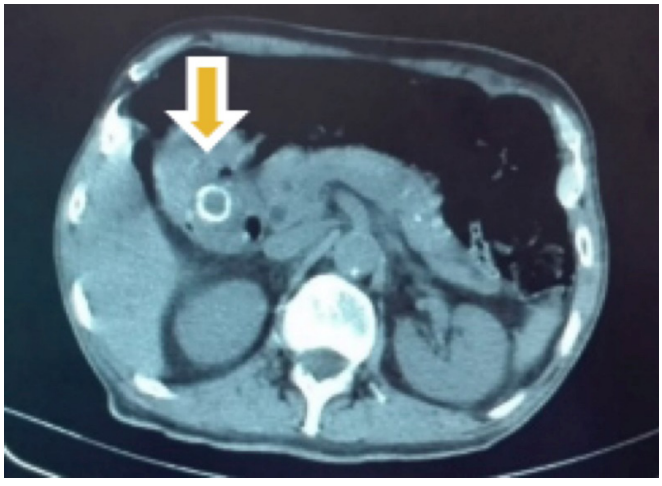

Figure 2 – **Ectopic gallstone in abdominal CT scan.**

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and a cholecysto-duodenal fistula. Moreover, pneumobilia in the left liver lobe with no dilation of the extrahepatic biliary tree was visible (figure 2).

Urgent laparotomy was performed. The gallstone was milked to the stomach and was removed by gastrotomy (figure 3). Due to unfavorable anatomic and physiologic conditions, cholecystectomy was not performed. Considering recurrence and operative risks an expecting approach was decided.

There were no complications in the perioperative period. The patient began feeding 5 days after surgery and remained asymptomatic. Regarding recurrence and operative risks, an expecting approach was preferred and one-year follow-up revealed no further symptoms.

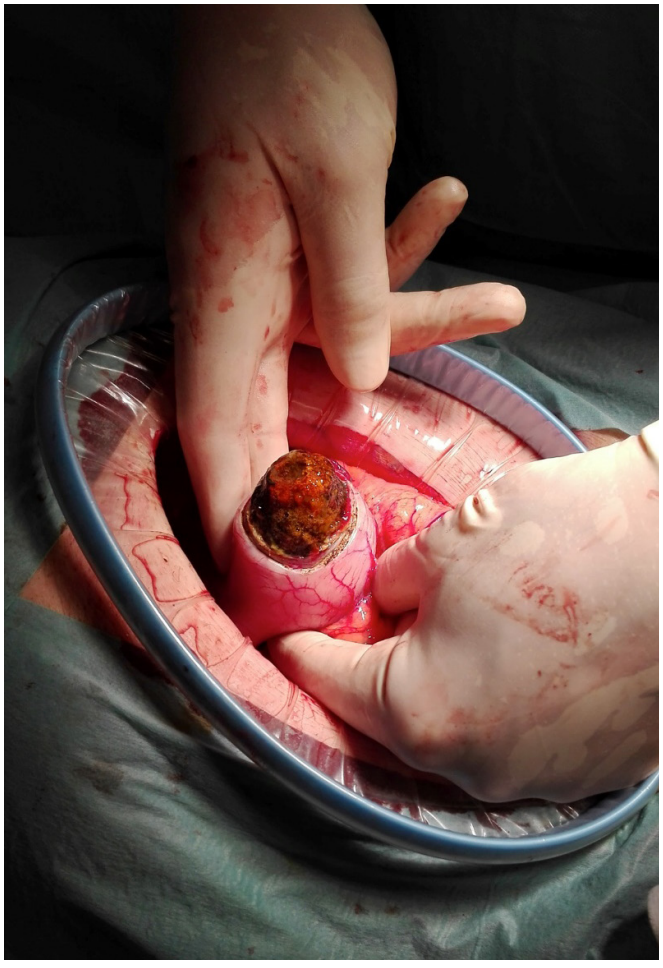

Figure 3 – **Gallstone removal by gastrotomy.**

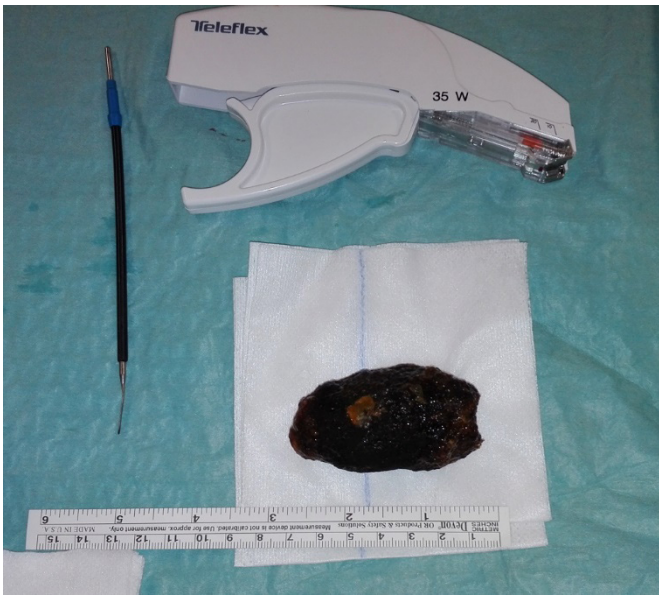

Figure 4 – **Gallstone with 3x6 cm.**

DISCUSSION

Bouveret's syndrome (BS) was first described in 1896 by Léon Bouveret.[4-6] It is defined as a gallstone ileus due to an acquired fistula between the gallbladder and either the duodenum or stomach. [2,3,5-8] BS often presents in elderly women with nonspecific symptoms (vomit, epigastric pain, anorexia).[6] There are described cases in which acute pancreatitis, gastrointestinal bleeding, duodenal perforation, Boerhaave's syndrome and gastric bezoars were the initial presentation.[4,5,7,8] The physical examination usually reveals abdominal distention, dehydration and hyperthermia.[6] Due to the patient's comorbidities and delayed diagnosis, morbidity can reach 60% and mortality 30%.[4]

It is essential a high index of clinical suspicion complemented with radiological findings. Rigler's triad (intestinal dilation, ectopic gallstone and pneumobilia) can be seen in plain abdominal X-Ray or abdominal sonography and is diagnostic in one third of the cases. CT-scan is diagnostic in 60% of cases, but only 25% of gallstones are isodense. Magnetic Resonance Imaging (MRI) should be used whenever possible in these cases. EGD is diagnostic

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in 70% of cases and can also be therapeutic, despite its low success rate (<10%).[3-7,9]

Although usually EDG is the first step, surgery is the definitive treatment in 91% of cases, with a mortality around 12%.[2,4,7] Gallstone fragmentation during EGD implies endoscopic removal of all pieces in order to avoid distal obstruction.[6-8] Surgical approach should be patient tailored, either by laparotomy or laparoscopy (elevated conversion rates) and guided by obstruction time.[3,10] Several authors recommend single removal of gallstone by enterotomy/gastrotomy in patients in shock or with relevant comorbidities. Although it remains controversial, cholecystectomy can be performed 4-6 weeks later if the patient remains symptomatic. In the other hand, in an otherwise healthy young patient cholecystectomy should be simultaneously performed with fistula treatment. Recurrent symptoms can appear in up to 56% of cases, especially if there are residual gallstones or obstruction of the cystic duct, with 10% of these patients requiring surgery. Furthermore, to date there seems to be no evidence that persistent cholecysto-duodenal fistula elevates the risk of developing gallbladder carcinoma.[2,3,5,7,8]

In this case report it is important to highlight the history of previous "gastric bezoar", symptoms of upper intestinal obstruction and radiologic identification of an ectopic gallstone. CT-scan was able to diagnose acute cholecystitis and to rule out the presence of more gallstones. Once EGD was not effective, the patient was submitted to an urgent laparotomy. The gallstone was removed by gastrotomy and no cholecystectomy was performed due to comorbidities. To date the patient remains asymptomatic, empowering our operative strategy.

CONCLUSION

BS is a rare complication of gallstone disease. It is often a delayed diagnosis specially in elderly women with comorbidities, which contributes to high morbidity and mortality. It is essential a high index of clinical suspicion complemented with radiological findings. Treatment can be either endoscopic or surgical and should be patient tailored. The most definitive procedures should be reserved for patients in the best physiological conditions.

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