CLINICAL CASE

Bouveret's syndrome: a rarest complication of cholelithiasis. A case report and literature review

Síndrome de Bouveret: una rara complicación de la colelitiasis. Informe de un caso y revisión de la bibliografía

Estephanía Navarro-del Río, and Jaime F. Hernández-Zúñiga*

Department of General Surgery, OPD Benemérito Hospital Civil de Guadalajara Fray Antonio Alcalde, Guadalajara, Jalisco, Mexico

Abstract

Bouveret's syndrome refers to the condition of gastric outlet obstruction caused by the impaction of a large gallstone into the duodenum after passage through a cholecystoduodenal fistula. Many endoscopic and surgical techniques have been described in the management of this syndrome; however, the morbidity and mortality are still very high. We present the case of a 67-year-old female patient with Bouveret's syndrome, with successful resolution with surgical treatment after two failed endoscopic treatments.

Key words: *Bouveret's syndrome. Gallstone ileus. Obstruction. Endoscopy. Surgery.*

Resumen

El síndrome de Bouveret se refiere a la obstrucción de la salida gástrica causada por un lito grande impactado en el duodeno que pasó a través de una fístula colecistoduodenal. Se han descrito varias técnicas endoscópicas y quirúrgicas para tratar esta entidad, pero la morbimortalidad es aún muy elevada. Se presenta el caso de una paciente femenina de 67 años con síndrome de Bouveret, con exitosa resolución mediante tratamiento quirúrgico posterior a dos tratamientos endoscópicos fallidos.

Palabras clave: *Síndrome de Bouveret. Íleo biliar. Obstrucción. Endoscopia. Cirugía.*

Introduction

Bouveret's syndrome is an uncommon form of gallstone ileus, caused by the passage and impaction of a large stone through a cholecystoduodenal fistula; the result is gastric outlet obstruction[1]. Beaussier first described it in 1770. In 1896, Frenchman Léon Bouveret published the first two cases of gastric outlet obstruction due to a stone impacted in the duodenal cap[2].

Since 2008, around 300 cases have been reported in the world literature. Morbidity and mortality of this syndrome have decreased in recent years but remain considerably high, estimated at 60% and 12-30%, respectively, depending on how timely diagnosis and treatment are[1].

Clinical case

Sixty-seven-year-old woman, with high blood pressure controlled with metoprolol and a 19-year history of type

Correspondence:
*Estephanía Navarro-del Río
Segunda Norte, 5172
C.P. 45030, Zapopan, Jalisco, México
E-mail: ssteph.navarro@gmail.com

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2 diabetes mellitus treated with NPH insulin (110 IU per day), poorly controlled, and with Grade II morbid obesity. She had a history of poliomyelitis with the lower limbs paraparesis as a sequel, and thus she was bed/chair-ridden. She attended the emergency department for intolerance to the oral route and weight loss. She referred her condition had started 20 days earlier with diarrhea (Bristol 7), with mucus, without blood, 10 bowel movements a day, with unquantified weight loss, intolerance to the oral route with coffee ground vomitus, more than 5 times a day, without pain; she attended her regional hospital, where gastroesophagoscopy revealed moderate erythema and duodenal cap with presence of a tumor lesion extending to the second portion of the duodenum at the fundus, body, and antrum of the stomach, which was the reason for her referral to the authors' institution. At admission to the emergency room, she had vital signs within normal parameters, and physical examination did not identify relevant data. Laboratory tests showed WHO Grade I hypochromic normocytic anemia, glucose at 174 mg/dL, albumin at 1.8 g/dL, and the rest within normal values. Abdominal computed tomography (CT) scan with intravenous contrast showed a collapsed gastric chamber due to the absence of gastric contents, a homogeneous-looking image with circumscribed edges of calcifying appearance to the left of the duodenum first and second portions, which generated duodenal compression, with the head of the pancreas pushed by the calcifying image (Fig. 1). Gastroendoscopy was performed (Fig. 2), where a stone of ~3.5 cm was recognized after the passage through the pylorus, obstructing 70% of the lumen and hindering the passage of the endoscope. A nasogastric bypass tube was placed, and she remained on fasting due to intolerance to the oral route. Total parenteral nutrition was started. During her approach, she developed an urinary tract infection and diabetic ketoacidosis, which resolved in 48 h. A new endoscopy was performed, whereby extracting a 2 cm × 1-cm stone from the second portion of the duodenum was possible, but a calculus remained in the third portion of the duodenum that could not be extracted; thus, an exploratory laparotomy was performed with the following findings: stomach adhered to the liver and scleroatrophic gallbladder with a fistulous tract toward the pylorus. A Kocher maneuver was performed, with a 3 cm × 3-cm stone being palpated in the third portion of the duodenum, as well as a 7 cm × 4-cm giant stone in the fourth portion. The fistulous tract was opened and the stones were extracted (Figs. 3 and 4). The pylorus was closed with the Heineke-Mikulicz technique. The bile duct could not be identified, and partial cholecystectomy was, therefore, performed. The patient had a good postoperative evolution, with oral route being initiated at 5th post-operative day. Gastroendoscopy was performed before discharge, where no fistulous tract was observed, but duodenal ulcers were identified in the cap and second portion of the duodenum (Forrest III). She was discharged on a proton pump inhibitor.

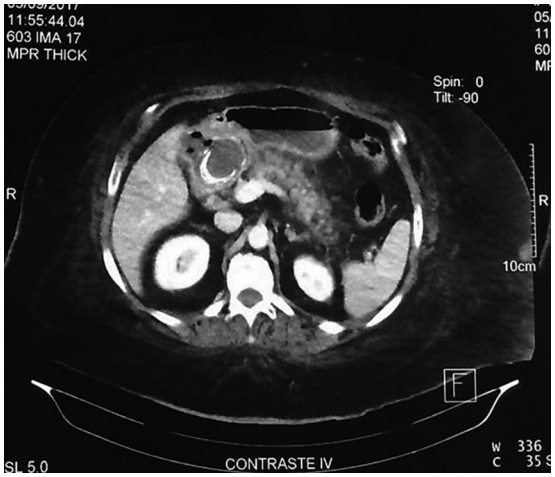

Figure 1. *Abdominal CT with IV contrast showing the gallbladder decreased in size, with gas within and compression due to increased duodenal volume; duodenal lumen showing a hypodense image with partially defined irregular dense wall. CT: computed tomography.*

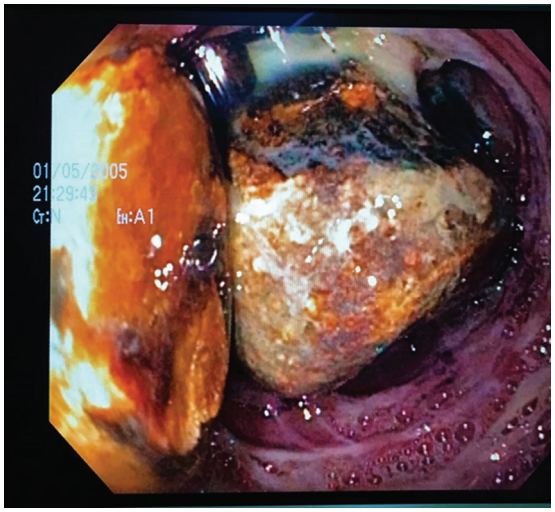

Figure 2. *Gastroendoscopy. After passage through the pylorus, a ~3.5 cm stone is observed that obstructs 70% of the lumen and hinders endoscope passage.*

Discussion

Biliodigestive fistula is a rare cholelithiasis complication that occurs in <1% of all patients. Fistula formation

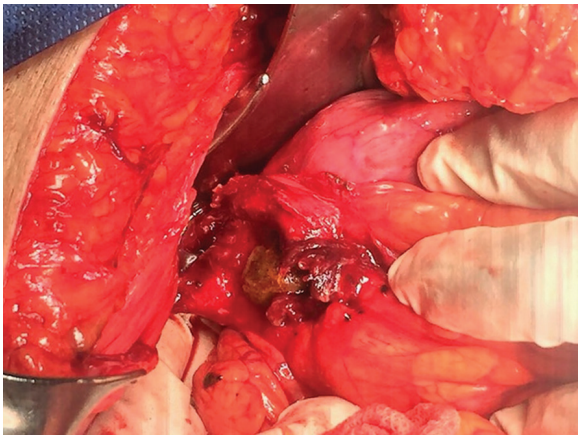

Figure 3. *Trans-surgical photograph; cholecystopyloric fistula with giant stone exiting is observed.*

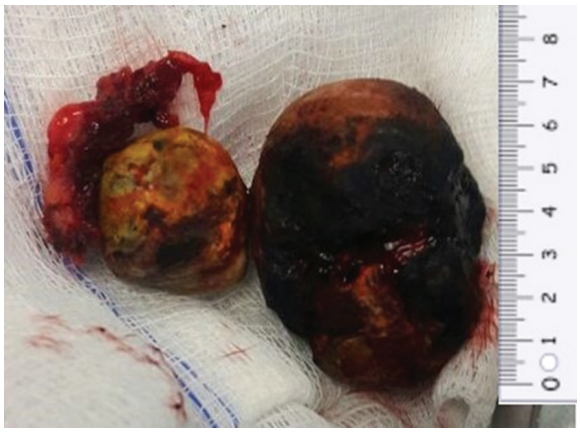

Figure 4. *3 cm × 3-cm and 7 cm × 4-cm giant stones extracted from the third and fourth portions of duodenum.*

is favored by a longstanding history of cholelithiasis, repeated episodes of acute cholecystitis, very large stones (2-8 cm), the female gender, and advanced age (more than 60 years). In 60% of cases (53-68%), the fistula is cholecystoduodenal[1].

The passage of stones through the fistula can trigger different clinical manifestations, depending on the size, the part of the compromised gastrointestinal tract and the previous existence of stenotic areas at this level. In 85% of all cases, the stone is expelled either by vomiting or feces. Conversely, in 15% of patients a gallstone ileus occurs[1]; this happens when a calculus of an adequate size passes through a cholecystoduodenal fistula or a choledochoduodenal fistula is impacted within the gastrointestinal tract, most often near the ileocecal junction, and distal obstruction of the small intestine occurs[2].

Gallstone ileus has been considered to appear in 0.3-0.4% of all patients with cholelithiasis and is seven times more common in patients older than 70 years. The most common localization, in 50-90% of cases, is the terminal ileum[1]. Stones smaller than 2.5 cm almost always pass through the small intestine, while larger stones remain impacted in the gastric outlet[3].

Bouveret's syndrome refers to the situation that occurs due to proximal impaction of the gallstone in the duodenum or distal stomach, which results in gastric emptying obstruction[1]. Léon Bouveret described it in two patients in 1896 and it is extremely rare. In general, 1-4% of intestinal obstructions result from gallstone ileus, and out of these, 1-3% are due to this syndrome[3].

Bouveret's syndrome most often affects elderly women with an average age of 68.6 years[1], such as this patient, with multiple comorbidities. Clinical presentation can be non-specific. Symptoms usually begin 5-7 days before seeking medical consultation. From 43% to 68% of patients have been reported to have a recent history of biliary colic, jaundice, or acute cholecystitis episodes, but cholelithiasis first manifestation as Bouveret's syndrome is also possible[1], as in the case of this patient, who was unaware of the presence of cholelithiasis and started with Bouveret's syndrome.

Nausea, vomiting, and abdominal pain at epigastrium and right hypochondrium are frequent, but intensity is hardly ever correlated with anatomical alterations. Fever, signs of dehydration, and weight loss may also be present. Sepsis is unusual. Bouveret's syndrome occurring with hematemesis, secondary to duodenal or celiac artery erosion, is even less common.

Given that clinical manifestations are typically nonspecific, in addition to patients' advanced age; it is a crucial situation that must be considered, as well as establishing a differential diagnosis of other causes of gastric emptying obstruction, such as gastric cancer and peptic stenosis, especially in elderly women[1]. This patient had melena and that was which led to requesting a gastroendoscopy at her first contact, whereby a neoplastic tumor was suspected before the correct diagnosis was established.

To determine Bouveret's syndrome diagnosis, Rigler's triad (pneumobilia, ectopic gallstones, and dilated stomach) is pathognomonic; however, in plain

abdominal radiograph, it is present only in 30-35% of cases[3].

The best imaging technique to identify Rigler's triad elements is abdominal CT. Furthermore, CT can demonstrate the biliodigestive fistula by contrast material or air, or it is indirectly suspected when the gallbladder is filled with oral contrast material; however, in 15-25% of patients, the stone cannot be visualized due to bile and fluid isoattenuation. In patients with intense emesis or intolerance to oral contrast, or in the case of isointense stones, the role of T2-weighted magnetic cholangioresonance can be important[1,2]. It can also be incidentally diagnosed by endoscopy[2].

Optimal treatment for patients with Bouveret's syndrome remains controversial in world publications. Therapeutic behavior should be planned after considering many parameters, such as patient general conditions, age, comorbidities, location of the stone, local inflammation, size of the calculus and fistula, and presence of more than one stone[1,4].

The primary goal of treatment is to resolve the obstruction by removing the impacted stone in the least invasive way possible[1]. Since it is an abnormality of elderly patients with comorbidities and high surgical risk, the endoscopic procedure is almost always the first line of treatment.

Despite many available endoscopic techniques, endoscopic treatment has a success rate of only 9%[3]. Another situation that should be considered is that fragmentation of the stones with endoscopic forceps can result in migration and impaction of the fragments in distal parts of the small intestine and lead to *de novo* intestinal obstruction. If endoscopy fails, surgery is the usual treatment[1].

By convention, Bouveret's syndrome has been treated with laparotomy and enterotomy as the simplest procedure, with relatively high morbidity and mortality, 60% and 30%, respectively[1,2]. Open, laparoscopic and manual laparoscopic procedures have been published with successful results in removing the impacted stone. Sica et al. reported the first laparoscopically-resolved Bouveret's syndrome case[4]. If possible, the stone should be moved to the stomach and be removed through a gastrostomy; if this is not possible, it can be moved to the small intestine and be removed by enterotomy[5]; the intention of these maneuvers is to make the incision on a healthy wall of the organ, which, potentially, heals adequately. If these two maneuvers fail, extraction should be performed through anterior duodenotomy, with special emphasis on optimal closure to avoid stenosis[1].

It is still a matter of controversy whether cholecystectomy and fistula repair should be performed in the same intervention[1]; many times it is difficult and is associated with a high risk of complications and it is often unnecessary, which is why many surgeons choose to perform a second operation only if the patient has symptoms[2]. Various studies comparing the combination of cholecystectomy and fistula closure with enterolithotomy alone indicate that simple stone removal is appropriate for most patients, with mortality rates of 20-30% and 12%, respectively[5]. Furthermore, a cholecystoduodenal fistula can function as a biliodigestive anastomosis if the biliary tract is permeable. Another argument supporting stone removal by enterotomy is that patients become asymptomatic after the procedure.

Gallstone ileus recurrence is rare[1]. In this case, the gallbladder was scleroatrophic and fistulized toward the pylorus, which is why opening the fistulous tract to remove the stones and performing partial cholecystectomy was decided (total cholecystectomy was not possible due to inflammation in the bile duct), as well as repairing the fistula with closure of the pylorus.

Post-operative mortality before the year 2000 was as high as 30%; however, early diagnosis by endoscopy and the advent of minimally-invasive techniques have made for mortality to decrease to 12%[2,3].

In the authors' institution, this is the first case of Bouveret's syndrome that has been operated in at least the last decade and, despite the poor general condition of this patient due to her multiple comorbidities; surgery was successful, without complications being observed at 1 year of post-operative follow-up.

Conflicts of interest

The authors declare that they have no conflicts of interest.

Ethical disclosure

Protection of people and animals. The authors declare that no experiments were performed on humans or animals for this research.

Confidentiality of data. The authors declare that they have followed the protocols of their work center on the publication of patient data.

Right to privacy and informed consent. The authors have obtained informed consent from the patients and/or subjects referred to in the article. This document is in the possession of the corresponding author.

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