Clinics in Surgery

Case Report
Published: 27 Jan, 2020

Subtotal Cholecystectomy with a Derivative Surgery as Treatment for Bouveret Syndrome

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Abstract

Bouveret syndrome is a rare complication of cholelithiasis; its usual clinical presentation is a gastric outlet obstruction. The absence of guidelines for the management of this condition, mainly because there are only few cases in the literature and the high clinical suspicion for its diagnosis, encourage us to elaborate this paper in which we review the diagnosis and treatment options of patients with this condition, presenting a report of one case in which we describe some of the endoscopic and surgical therapies used.

Keywords: Bouveret syndrome; Cholecysto-duodenal fistula; Gallstone ileus; Obstruction; Endoscopy; Subtotal Cholecystectomy

Introduction

First described by Beaussier (1770) and published 100 years after by L. Bouveret with 2 cases (1896), Bouveret´s syndrome is a very rare type of gallstone ileus caused by the passage and impaction of a large gallstone through a cholecystoduodenal/gastrocholecystic fistula into the duodenum or stomach, resulting on a gastric outlet obstruction eventually [1-3]. Complications associated with cholelithiasis are common and include acute cholecystitis, choledocolithiasis, pancreatitis, and gallstone ileus, in which Bouveret syndrome is the rarest example. Gallstone ileus is responsible for 1% to 4% of all cases with small-bowel obstruction [2]. Gallstone ileus is extremely rare, appearing as a complication only 0.3% to 0.5% of patients with cholelithiasis. The most common location of a calculus causing obstruction is the terminal ileum. More proximal impaction is unusual whereas obstruction of the duodenum accounts for 1% to 3% of all cases. Bouveret syndrome represents ~1% to 3% of cases of gallstone ileus [2-6]. Due to the rare nature of this disease, there are no standardized recommendations for the diagnostic workup and management of these patients, including endoscopic, laparoscopic, and open surgical options. There has been reported among 300 cases reported in the world literature, and the rates of morbidity and mortality has been decreasing from 60% to 20%-30% these days, but still remaining high due to the advanced age and comorbidity of the patients [3-6]. This rate will depend on the therapeutic approach almost strictly.

Case and Methods

A 65 years old patient with a past of atrial fibrillation, ischemic cardiopathy with cardiac infarction with coronary arteries stents, diabetes, high blood pressure, Crohn disease with no acute events lately, gallbladder and kidney stones and a hepatocarcinoma stage IV with bone metastasis came to the urgency unit with vomits, abdominal pain and incapability of swallowing food completely (Figure 1). Physical examination showed mild pain in the upper abdomen with some bloating. He had been discharged from the gastroenterology unit a week before with a diagnosis of chronic cholecystitis with amoxicillin/clavulanic acid oral treatment which he couldn't finish because of the incoming clinical event (Figure 2). During his hospitalization in our surgical unit we ran some tests. Most important were a TC scan (with oral and IV contrast) from the urgency in which it's suggested a cholecystoduodenal fistula because of the presence of a large stone in the gallbladder and another large one (2.5 cm × 2.5 cm) in the 1st duodenal portion and gas bubbles inside the gallbladder (Figure 3). All of this was also confirmed with a cholangio-MRI. In neither of those studies a clear fistulous channel was identified (although the oral contrast also was used). Both imaging tests also mentioned the already documented 4B segment hepatocarcinoma. On a gastroduodenoscopy (EDG) done afterwards, a large stone of 2.5 cm of diameter was identified but unable to be taken out because it was impacted in the 1st duodenal portion (Figure 4). Being run out almost all therapeutic options, an in order to prevent any complications that may occur if a

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Received Date: *23 Dec 2019*
Accepted Date: *21 Jan 2020*
Published Date: *27 Jan 2020*

Citation:
Villachica JT, De La Guardia Rueda C, Teillet Roldán MÁ, Fernández MA. Subtotal Cholecystectomy with a Derivative Surgery as Treatment for Bouveret Syndrome. Clin Surg. 2020; 5: 2717.

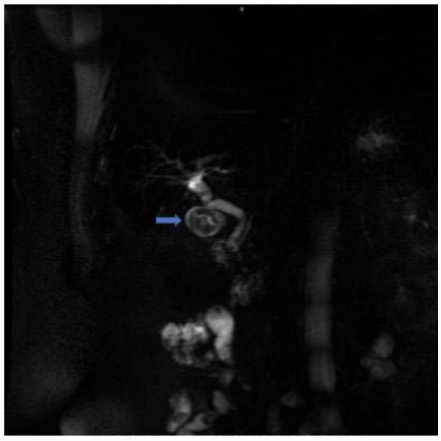

Figure 1: Cholangio-MRI of the patient showing a stone in the gallbladder.

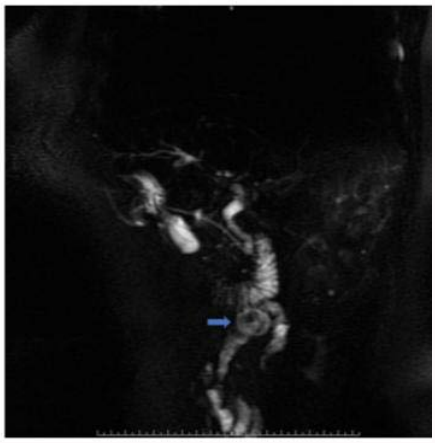

Figure 2: Same Cholangio-MRI showing another gallstone in the proximal duodenum.

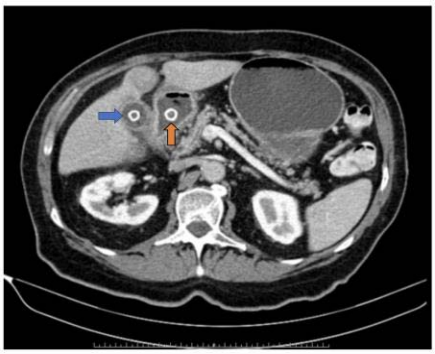

Figure 3: Blue arrow: Gallbladder with a large stone with gas bubbles around. Orange arrow: Gallstone located in the upper small bowel segment. Dilated stomach with a vast content of fluid and gas.

treatment is not set in time, we decided surgery was the only available solution for this case despite the number of comorbidities the patient had. Shortly before the surgery another TC scan confirmed the absence of stones in all the gastrointestinal system (present only in the gallbladder), also by then the patient got an important clinical improvement (Figure 5). Either way we still programmed the surgery despite this improvement because of the risk of recurrence of bowel obstruction with new and more complicated episodes.

Results

We proceed to do an open approach by a right subcostal incision extended to the left quadrant. An intraoperative manually guided endoscopy was done confirming the absence of the gallstone in the entire upper Gastrointestinal Tract (GIT) (until the last portion of the duodenum). These findings were confirmed before on a TC scan with oral and IV contrast. Then we proceed to do a subtotal cholecystectomy leaving the gallbladder infundibulum sutured an attached to an omentum patch because we were unable to clearly identify and separate the main bile duct from the gallbladder hilum and also to prevent damages to these structures and its surroundings. We also performed a gastro-jejunal anastomosis with mechanical section and closure of the duodenum (choosing a healthy, not inflamed tissue) and an Y de Roux entero-enteric anastomosis. Patient was discharge after 2 weeks of postoperative with no complications of the surgery, clinically he presented just a minor epigastralgia that was well managed with oral PBI (pantoprazole) and sucralfate having good results being asymptomatic in consultation one month later his discharge. A postoperative (PO) TC scan didn't show any bowel obstruction recurrences or complications of the surgery. Patient could retake his chemotherapy treatment soon after this first PO consultation.

Discussion

This clinical condition remains difficult to diagnose as the symptoms are nonspecific and the physical exam findings may be very subtle. The key to diagnosing patients with Bouveret´s syndrome is maintaining a high level of suspicion in elders with a history of cholelithiasis and symptoms of gastric outlet obstruction. The largest review (128 cases) of this syndrome found that over 85% of clinical presentation was with nausea and vomiting, abdominal tenderness could appear among 70% of them, less common GI bleeding (21%). Mainly these patients with clinical presentation bowel obstruction will get the suspicion of this syndrome when the following findings appear on an X- Ray test: pneumobilia, a bloated stomach with an enteric gallstone suggested image (Rigler's triad). Usually if the pain is in the right upper quadrant the diagnose could be given on an ultrasound test showing thickening of the gallbladder wall with or without cholecystitis [2,7]. More extended and detailed tests like a TC scan and a MRI would demonstrate better the pneumobilia, dilated stomach and gallbladder stones in the GI system also using and oral contrast which would demonstrate extravasation of contrast into the gallbladder. The acknowledgement of a fistula or pneumobilia will be present in the majority of cases and gallstones on half of them. In our case pneumobilia, a large stone in the gallbladder with another large one in the 1st duodenal segment (that probably migrated through the cholecystoduodenal fistula) and a highly dilated stomach confirmed the suspected diagnosis. Endoscopy is both diagnostic and potentially therapeutic and since this syndrome would happen on elderly comorbid patients, many specialists are choosing endoscopy as first-line therapy. Multiple endoscopic modalities have been utilized for treating Bouveret syndrome (endoscopic nets/baskets, mechanical, electrohydraulic, laser, and extracorporeal shockwave lithotripsy) as well as combined endoscopic and surgical therapies. Stones in the GI tract may be seen in two-thirds of the cases and the proper extraction of them is even less probable to happen [2,9-11]. The visualization of the fistula (whether if it is cholecystogastric or cholecystoduodenal) is difficult and even more its closure which is not accomplished in most of the cases and typically not necessary

Figure 4: Preoperative EDG showing a large (2.5 cm × 2.5 cm) impacted gallstone in the duodenal bulbus unable to be extracted by this procedure because of technical difficulties and high risk of perforation.

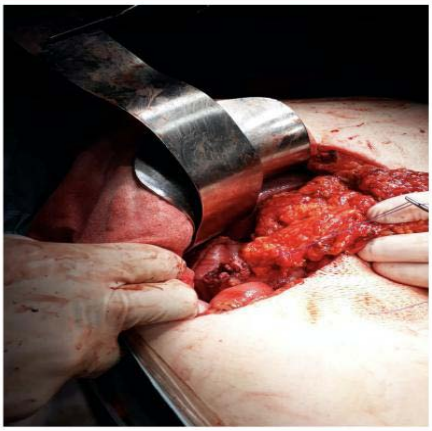

Figure 5: Suture of the gallbladder infundibulum after performing the subtotal cholecystectomy.

Figure 6: Suture of the omentum patch above covering the gallbladder infundibulum.

[2,8-11]. In our case the preoperative endoscopy was able to identify the gallbladder stone in the duodenal bulbus but failed to see the fistula and extract the stone even though it was teared into smaller pieces, because it was impacted to its mucosa. Gallstones can migrate after this procedure and compromise the patient with a more distal ileus, that is why we followed this patient very closely taking blood samples and imaging tests in which no signs of complication where shown, yet an improvement of clinical markers was observed. TC scan and EGD are preferred in Bouverets syndrome cases because of their highest sensibility and specificity among others [12-14]. Surgery must be considered in cases that endoscopy fails, or the clinical condition of the patient worsens. In cases in which stones can be maneuvered with low risk of disruption of the bowel wall (extensive ulceration of the mucosa) a gastrostomy, pylorotomy or duodenotomy at or immediately proximal to the site of obstruction are advised [2,8,11,12]. If the stone migrate into the distal duodenum or the proximal jejunum an enterotomy could be done distal to the Treitz ligament and an examination of all the small bowel should be concluded in order to avoid further postoperative obstruction by remnant stones [2,13]. In our patient we developed an open surgical approach; an intraoperative endoscopy guided manually was done to confirm the absence of the gallbladder stones in the upper GIT (confirmed preoperatively on a TC scan with oral and IV contrast). We proceed to do a subtotal cholecystectomy leaving the gallbladder infundibulum sutured an attached to an omentum patch because we were unable to identify clearly and separate the main bile duct from the gallbladder hilum and to prevent lesions around these structures (a duodenotomy and further suture on this place with local fibrosis and swelling might leak), we also did a gastro-jejunal anastomosis with mechanical closure of the duodenum (healthy, not inflamed tissue) an Y de Roux entero-enteric anastomosis to prevent compression on the site of the duodenal fistula avoiding further leakage of bile or enteric content.

Conclusion

Bouveret syndrome is a very unusual clinical presentation of patients with gallstones which demands a high level of suspicion for its diagnosis and this begins usually with routine imaging testing associated with gastric outlet obstruction symptoms. We also like to emphasize the importance of endoscopic diagnostics and therapeutics properties on this matter because the common patient that suffers this condition is old with other much comorbidity and will benefit with a non-aggressive procedures like the endoscopy, although the location and closure of the fistula is rarely possible. When endoscopic treatment fails, the recommended procedure is an incision at, proximal or distal to the location of the gallstone stuck in the small bowel. We present this case as a reminder that surgery could be the only alternative in many centers on this matter, could it be because of failure of the endoscopic approach or the lack of other therapies (mechanical, electrohydraulic, laser, and extracorporeal shockwave lithotripsy) so we should be prepared to provide a safe option to the patient. Although cholecystectomy and the treatment of the fistula is generally not required, only in special cases, we think this case is an useful example of this because many cases of Bouveret syndrome are associated with cholecystitis and swelling/fibrosis of the structures near the gallbladder's infundibulum getting us into surgical technical difficulties requiring us to develop as it happened in this case, a subtotal cholecystectomy with a gastro-jejunal bypass. We think in situations that is not possible to treat the patient endoscopically nor perform an enterotomy/gastrotomy, this procedure is an optimal alternative

because it will cut the passage of gallstones to the small bowel and prevent further obstruction events by doing the cholecystectomy and prevent a harmful and risky event like an injury of the main bile duct and its surroundings (subtotal cholecystectomy with omentum patch) preventing a bile-enteric leakage by decompressing the duodenal fistula site (gastro-jejunal bypass).

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