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Review

Bouveret's Syndrome: A Case-Based Review, Clinical Presentation, Diagnostics and Treatment Approaches

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Abstract

Gastric outlet obstruction that arises from gallstones impacted in the distal stomach or proximal duodenum after passing through a cholecystoduodenal, cholecystogastric or rarely choledocoduodenal fistula is called as Bouveret's syndrome and it accounts for approximately 1-3% of all the patients with gallstone ileus. Although treatment modalities, including stone removal or fragmentation with classical endoscopic devices, such as snares, and forceps or fragmentation of gallstones with new devices, such as electrohydraulic lithotripsy, laser, extracorporeal shockwave lithotripsy have been described. However, only 29% of the patients benefit from nonsurgical methods. Removal of the stone through a gastrotomy or enterotomy and performing cholecystectomy and fistula repair with a second operation is an approach recommended for older patients with comorbid diseases. In this paper, a case of Bouveret's syndrome was presented. The authors also aimed to review the diagnosis, management and treatment of this rare disease and to update the previous reviews.

Keywords: Bouveret's syndrome; gallstone; ileus.

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Complications related to gallstones are not rare in daily medical practice, which are acute cholecystitis, pancreatitis, choledocolithiasis and gallstone ileus. Gallstone ileus is obstruction of the gastrointestinal system that arises from gallstones and occurs only approximately 0.3-0.5% of the patients having gallstones.[1] Obstruction usually occurs in the terminal ileum and ileocecal valve (70%), rarely occurs in proximal ileum or jejunum (20-40%).[2] Gastric outlet obstruction that arises from gallstones impacted in the distal stomach or proximal duodenum after passing through a cholecystoduodenal, cholecystogastric or rarely choledocoduodenal fistula is called as Bouveret's syndrome (BS). The first two cases were described for the first time in 1896 by a Parisian surgeon, Leon Bouveret.[3] This rare complication is observed only 1-3% of patients having gallstone ileus and more frequent in patients with advanced age and comorbid diseases.[4] It is mostly presented as single case reports; the largest case series in the literature only consist of six cases.[5] Because of the rarity of this disease, no standardized diagnostic or therapeutic technique is described, including endoscopic techniques or surgical approaches (open or laparoscopic). In this paper, a case of BS was presented. The authors also aimed to review the diagnosis, management and treatment of this rare disease and to update the previous reviews. The patient's written consent was obtained in this paper.

Methods and Case Presentation

Keywords "Bouveret", Bouveret syndrome", "Bouveret's" and "Bouveret's syndrome" were searched in PubMed and

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Google Scholar database. All related articles published in English between the years 1968-2018 were evaluated. The 152 cases were found in 146 articles. Gender, age, symptoms, signs, diagnostic tools, treatment options were analysed. Also, a new case in our clinic was presented below.

A 69-year-old female patient was evaluated due to complaints of loss of appetite, nausea began 10 days ago and complaints of vomiting and abdominal pain after eating began two days before the patient was admitted to our hospital. On examination, the blood pressure of the patient who was on salbutamol 200 mcg, valsartan 320 mg, hydrochlorothiazide 25 mg treatment due to hypertension and chronic obstructive pulmonary disease was 155/90 mmHg. Her heart rate was 88/bpm, and her body temperature was 37.8ºC. She also had a history of type 2 diabetes mellitus under insulin treatment. On physical examination of the abdomen, she had tenderness in her right upper quadrant, and her intestinal sounds were hypoactive. At laboratory examinations, leukocyte was 12300/mm^3(4600-10200/mm^3), c-reactive protein was 27 mg/dL (0-5 mg/dL) and potassium was 2.8 mmol/dL (3.5-5.1 mmol/dL) aspartate aminotransferase was 154U/L (5-34 U/L), alanine aminotransferase was 205 U/L (0-55 U/L), and she had a slightly elevated alkaline phosphatase level of 177 U/L (40-150 U/L). In her blood gases analysis, she had a pH of 7.31. Abdominal x-ray showed no abnormal findings. Abdominal ultrasound examination revealed an image in size of 4 cm with acoustic shadowing in the duodenum. Computed tomography scan with intravenous and oral contrast of the abdomen demonstrated that the stomach was dilated, and there was a solid mass approximately in size of 4 cm in the duodenal bulb (Fig. 1). In consequence of gastroscopy performed, a gallstone impacted the duodenal bulb in size of 4 cm was observed (Fig. 2). Gallstone was tried to be split by using endoscopic forceps and snares, but since this procedure failed, it was decided to perform surgery. In exploration performed with a supraumbilical vertical midline incision, it was observed that the gallbladder, duodenum and surrounding fatty tissues were oedematous; the areas of inflammation were wrapped by the omentum. Because the wall of the duodenum is very oedematous and fragile, it was decided to reach the stones via gastrotomy. After performing gastrotomy from the pyloric area, the impacted gallstone has been extracted in two pieces through gastrotomy using sponge forceps, and subsequent Heineke-Mikulicz pyloroplasty was performed. A 7-mm silicone drain was inserted in the operation area. And after closing the abdominal midline incision, the operation was finished uneventfully. Since our patient's advanced age, poor general condition and her comorbid diseases, cholecystectomy was not performed in the same operative session. After surgery, the patient's c-reactive protein level became normal on the 6th day of surgery. On the fourth day of the surgery, the liquid diet was given to the patient and she was discharged on the postoperative 7th day uneventfully. Since she did not come for follow-ups after discharge, cholecystectomy in the second session could not be performed.

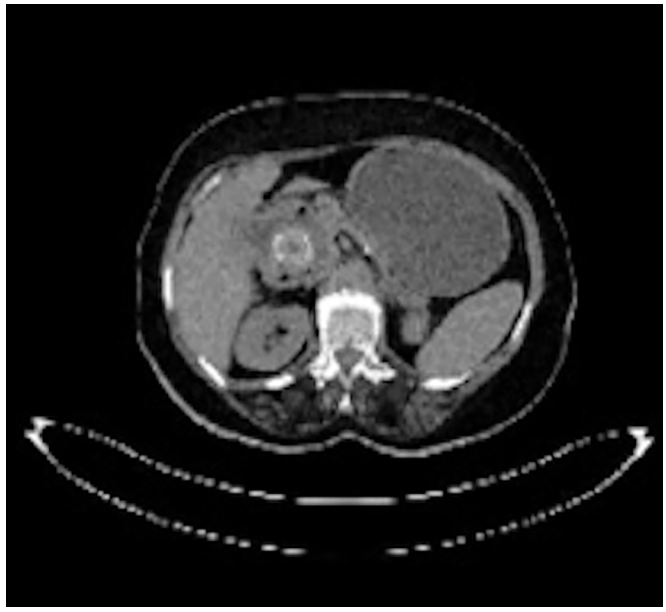

Figure 1. Gallstone impacted in the duodenum (blue arrow) and dilated stomach stone (white arrow) on tomography.

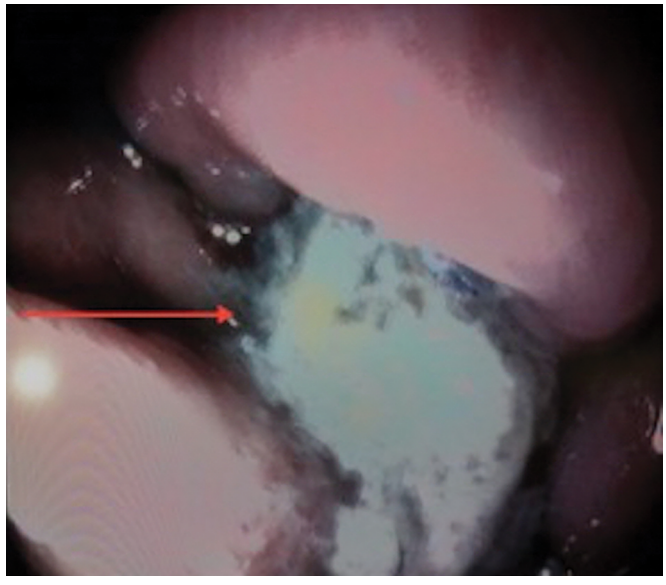

Figure 2. Gallstone impacted in the duodenal bulb (red arrow).

Discussion

Fistulas between the biliary system and the upper gastrointestinal system are rare pathologies, but BS is an even rarer

phenomenon that has the potential to cause lethal complications. BS tends to occur more commonly in women (65%) with a median age of 74.1, and patients mostly have comorbid diseases.[4] A higher incidence of BS in females can be explained by the higher rates of gallstones in females than males, which is related to cholestatic effects of oestrogen and progesterone.[6] Ileus, due to gallstones, has female predominance related to the same factors. Advanced age and gender of our patient are typical for BS.

The size of gallstones is an important risk factor for BS. Large gallstones (>2.5 cm) have the potential to migrate into the duodenal bulb or distal part of the stomach, where they frequently lead to obstruction by eroding gallbladder or choledochal wall.[7] The gallstone of the presented case was 4 cm in size, which is typical for gallstone ileus.

BS has nonspecific symptoms, physical and radiological findings. Knowledge of risk factors, signs, symptoms, radiological and endoscopic findings are important for rapid and accurate diagnosis. However, the most important key to diagnosing is having a high level of suspicion in cases with a history of gallstone and sings of pyloric obstruction. The disease mimics pyloric obstruction very well. It may cause nausea, vomiting, abdominal pain or discomfort, loss of appetite, weight loss and anorexia. Symptoms of dehydration also may be observed due to vomiting. Hematemesis or melena may develop respectively, 13.8% and 4.6% of BS cases due to mucosal erosion that arises from an impacted gallstone.[1] In recently reported 152 cases, vomiting, nausea and abdominal pain were the most common symptoms (Table 1). Our patient presented all three symptoms.

Abdominal tenderness, distension due to obstruction of the distal stomach or duodenal bulb and absence or hypoactivation of intestinal sounds is the most observed signs (Table 2). Our patient presented all three major signs.

Table 1. Symptoms of the patients with the Bouveret's syndrome (A review of 152 cases in 138 reports)

Symptoms	Number of the findings	% of the findings
Nausea/Vomiting	124	81.5
Abdominal pain	103	67.7
Hematemesis	21	13.8
Weight loss	18	11.8
Anorexia	17	11.1
Constipation	11	7.2
Melena	7	4.6
Pale stool	2	1.3
Diarrhea	3	1.9
Dysphagia	2	

Since the clinical presentations were not reported, three of 152 cases were excluded (Reference number 44, 45, 46 were excluded)

Table 2. Sings of the patients with the Bouveret's syndrome (A review of 152 cases in 138 reports)

Sings	Number of the findings	% of the findings
Abdominal tenderness	59	38.8
Abdominal distention	39	25.6
Hypoactive or absent of bowel sounds	7	4.6
Jaundie	3	1.97
Succussion splash	10	0.65

Since the clinical presentations were not reported, three of 152 cases were excluded (Reference number 44, 45, 46 were excluded)

Metabolic alkalosis, hypokalaemia secondary to severe vomiting and elevation in leukocyte and CRP values due to the inflammatory process in the gallbladder, fistula and surrounding tissues can be seen in patients. Our patient had an increase in CRP value over 50-fold and leucocytosis. Additionally, hypokalaemia was determined.

The following findings can be observed on an abdominal x-ray or upper gastrointestinal system fluoroscopy: intestinal obstruction, gallstone in the intestinal lumen and the presence of gas in the biliary system (pneumobilia). These findings are known as Rigler's triad. In addition to these, Balthazar and Schechter also added two findings, air-fluid levels in the right upper quadrant and air in the gallbladder and duodenal bulb on abdominal x-ray.[8] However, this triad of x-ray findings is observed only in 30-35% of the cases (Table 3) (7,9). Since obstruction is in the proximal part in BS, classical gallstone ileus findings cannot be observed in the abdominal x-ray. In the presented case, no significant pathology was determined in the abdominal x-ray.

Ultrasonography and computed tomography are helpful for diagnosis; these non-invasive tests are preferred as confirmation of endoscopic diagnosis. Ultrasonography is useful in identifying gallstones in the gallbladder. It has limited ability to identify stones in duodenum or stomach

Table 3. Abdominal X-ray findings in 81 cases of the Bouveret's syndrome

Findings	Number of the findings	% of the findings
Pneumobilia	30	37
Calcified mass or gallstone	29	35.8
Dilated stomach	17	20.9
Dilated intestinal loops	10	12.3
Paucity of bowel gas	2	2.5
No significant findings	11	13.5

The 81 cases of BS from references: 4-6, 17, 20, 28, 29-32

compared to computed tomography [9] (Table 4). In our presented case, ultrasonography identified gallstone in the duodenal bulb. Computed tomography is helpful in identifying fistula between gallbladder and stomach or duodenum, also helpful to show the accurate size of the impacted stone. However, diagnosis can be made by using computed tomography in 60% of patients.[8] Computed tomography also cannot show choledocus lumen clearly in 25% of the cases. However, magnetic resonance cholangiopancreatography clearly shows the whole biliary system and differentiates fluid from calculi.[10, 11] Magnetic resonance cholangiopancreatography also identifies fistula between the biliary system and the gastrointestinal system.[10] It can be used for evaluation if the pericholecystic fluid is found or not and evaluation of gallbladder, stomach and the intestinal wall.[12, 13] Oral contrast usage in computed tomography or magnetic resonance imaging demonstrates contrast extravasation into gallbladder via the fistula. Analysis of 67 computed topographies of BS patients revealed that the fistula and pneumobilia diagnosis rate is 46.2% in computed tomography (Table 5). Despite all developments in diagnostic techniques, the diagnosis can be made only in 50% of the cases preoperatively.[14]

Table 4. Ultrasonography findings in 48 cases of the Bouveret's syndrome

Findings	Number of the findings	% of the findings
Gallstone in gallbladder	24	50
Gallstone in duodenum	10	20.8
Thickened gallbladder wall	4	8.3
Unspecified echogenic focus in right upper quadrant	2	4.1
Cholecystoduodenal fistula and pneumobilia	18	37.5
No significant findings	6	12.5

The 48 cases of BS from references: 4, 5, 6, 25, 30, 47

Table 5. Computed tomography findings in 67 cases of the Bouveret's syndrome

Findings	Number of the findings	% of the findings
Cholecystoduodenal fistula or pneumobilia	31	46.2
Gallstone in duodenum	24	35.8
Gallstone in gallbladder	13	19.4
Gallstone in stomach	3	4.4
Mass in duodenum	15	22.3
Mass in pylorus	1	1.5
Mass in gallbladder	1	1.5
Dilatation in stomach	12	17.9
Dilatation in duodenum	7	10.4

The 67 cases of BS from references: 1, 3-6, 22, 23, 28, 29-39, 42, 43, 47, 48

Table 6. Endoscopic findings in 90 cases of Bouveret's syndrome

Findings	Number of the findings	% of the findings
Dilated stomach	4	4.4
Obstruction		
Pylorus	12	13.3
Duodenal bulb	59	65.5
Postbulber duodenum	19	21.1
Fistula		
With stone	4	4.4
Without stone	7	7.7
No significant findings	2	2.2

The 90 cases of BS from references: 1, 4, 6, 20, 22-24, 34, 36, 37, 39-43, 47*
*ERCP was performed

The upper gastrointestinal system endoscopy shows the impacted stone in the distal stomach or duodenal bulb with a dilated stomach (Table 6). The duodenal orifice of the cholecystoduodenal or choledocoduodenal fistula may also be identified in upper gastrointestinal system endoscopy.[15-17] Although treatment modalities, including stone removal with endoscopic techniques (nets/baskets) or fragmentation of gallstones with mechanical, electrohydraulic, laser or extracorporeal shockwave lithotripsy, have been described, only 29% of the patients benefit from endoscopic methods.[18] Mechanical lithotripsy can be used to split stones into small pieces before extracting impacted stone by nets or baskets. Baskets, snares, forceps may be used as mechanical lithotripter.[19] If suspicion of malignancy occurs, biopsies should be taken at the time of the endoscopic approach. Extracting all stone pieces is necessary to avoid postoperative ileus due to gallstones.[20]

Electrohydraulic lithotripsy is another option as an endoscopic treatment modality. The first usage of electrohydraulic lithotripsy in endoscopy is described by Sethi et al. They used a double-channel gastroscope during the operation.[21] This method may have the potential of complications as perforation and bleeding due to damage in surrounding tissues.

Neodymium, rhodamine, holmium: YAG lasers are also promising techniques for gallstone fragmentation. The advantage of laser using is splitting the stone with minimal tissue damage.[22, 23] Holmium: YAG laser fragmentation has benefits over neodymium and rhodamine laser given the capability of applying high energy via flexible and small probes.[24]

Holl et al. first reported the success of extracorporeal shockwave lithotripsy in gallstone impacted in duodenum.[25] After Holl et al., many cases were reported about extracorporeal shockwave lithotripsy treatment. Most of these cases have shown to be ineffective or addition of other endoscopic techniques required. In a case reported by Lenz et al., five times of extracorporeal shockwave lithotripsy was used to split the stone, which impacted in the duodenal bulb.[26]

Limitation of endoscopic therapy is the impossibility of closure of fistula between stomach/duodenum and gallbladder or choledocus. Another limitation is defragmented stones may cause gallstone ileus by obstructing distal parts of intestines. Beside all these limitations, endoscopic treatment methods need a high degree of experience. Endoscopic treatment methods may be insufficient for the stones that are larger than 2.5 cm.[4] An attempt of endoscopic treatment for the stone in size of 4 cm determined in our case failed.

As the surgical treatment modality, removal of the stone through a gastrotomy or enterotomy and in addition to this operation performing cholecystectomy and fistula repair was described. Although the surgical approach is the most efficient method, due to comorbid factors in most of the patients, postoperative mortality rates may reach a level of 12%; in fact, when cholecystectomy is added, these rates may increase much more.[27] Removal of the stone through a gastrotomy or enterotomy and performing cholecystectomy and fistula repair in a second operation is an approach recommended for elderly patients with comorbid diseases (Table 7). The likelihood of occurrence of gallstone ileus before the second operation is 5%.[28] Cholangitis and cholecystitis are other complications that may occur before the second operation. Since our patients advanced age, poor general condition and her comorbid diseases, cholecystectomy was not performed in the same operative session.

Conclusion

Gallstone ileus is an infrequent complication of gallstone disease, but BS is a scarce complication of gallstone disease. BS presents differently from classical gallstone ileus with symptoms and signs of gastric outlet obstruction. Recognizing risks, symptoms, sings and radiological findings that are pointed out in this review is important for early diagnosis. Gallstone disease, ileus and BS have a higher incidence in the female population due to cholestatic effects of oestrogens. Gender and advanced age of this reported patient is typical for this rare disease. Small gallstones pass through the cystic duct, whereas large gallstones cannot pass and erode gallbladder wall and lead to a fistula between gallbladder and duodenum. In recent years, diagnostic tools and treatment strategies evolve to less invasive methods.

Table 7. Treatment modalities and clinical outcomes of 129 patients of Bouveret's syndrome

Treatment modality	Cases	Success number	Mortality	Major complication
Endoscopic extraction	39	4	0	0
Laser lithotripsy	7	5	0	0
ESWL	5	1	0	0
Mechanical or electrohydraulic lithotripsy	16	3	0	1 e.coli sepsis
Enterotomy/gastrotomy+stone extraction	49	45	1	1 respiratory failure, 1 wound dehiscence,1 duodenal leak, 1 fungal sepsis
Enterotomy+stone extraction+cholecystectomy+fistula repair	36	30	1	1 sepsis, 1 renal failure, 1 upper GI bleed, 1 pulmonary embolism, 1 bile leak
Enterotomy+stone extraction+cholecystectomy	3	2	0	1 bile leak
Enterotomy+stone extraction+fistula repair	3	3	0	0
Enterotomy+stone extraction+cholecystectomy+pyloroplasty	3	3	0	0
Antrectomt+gastrojejunostomy+cholecystectomy	1	1	1	0
Laparoscopic enterolithotomy	2	2	0	0
No therapy	1	0	1	0

Since the outcome of the case was not reported, the 23 of 152 BS cases were excluded from this Table (Excluded references: 5, 37, 40, 42, 48)

The use of laser technology with endoscopic tools promises a new era in the treatment of this rare disease. Avoiding surgical intervention may decrease mortality in cases with comorbid diseases; endoscopic interventions may be tried in these cases. However, in cases having the necessity of surgery, two-stage surgical interventions should be preferred in patients with comorbid risk factors.

Disclosures

Ethics Committee Approval: The study was approved by the Ethics Comitee of Istanbul Okan University Hospital (09.05.2018/94).

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Conflict of Interest: None declared.

Authorship Contributions: Concept – M.F.F.; Design – M.F.F.; Supervision – A.K.; Materials – M.F.F.; Data collection &/or processing – M.F.F., A.K.; Analysis and/or interpretation – M.F.F.; Literature search – M.F.F., A.K.; Writing – M.F.F., A.K.; Critical review – M.F.F., A.K.

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