

Endoscopic versus surgical treatment for a rare form of gallstone ileus: Bouveret's syndrome.

Two case reports and review of the literature



Ann. Ital. Chir., 2018 89, 2: 162-167
pii: S0003469X18027471

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Endoscopic versus surgical treatment for a rare form of gallstone ileus: Bouveret's syndrome. Two case reports and review of the literature.

BACKGROUND: *Bouveret's syndrome is a gastric outlet obstruction due to a gallstone impacted in the duodenal lumen or in the stomach via a bilioenteric fistula. It's the rarest form of gallstone ileus, with few cases reported in medical literature. Typically, this condition affects elderly people and causes high morbidity and mortality rates.*

METHODS: *We present a review of the published cases of Bouveret's syndrome between 2006, year of publication of the largest case series, and 2015 with particular regard to the operative management and treatment options. Two demanding cases of Bouveret's syndrome reported in our Hospital are also described.*

RESULTS: *Bouveret's syndrome is a rare clinical condition and data reported in medical literature only derive from single case reports and case series. Therefore no clear management indications have been ruled out and treatment options are still matter of debate.*

CONCLUSION: *Bouveret's syndrome is a rare clinical entity with a challenging operative management. Surgical or endoscopic treatments should be tailored on patient medical conditions and clinical presentation.*

KEY WORDS: Bouveret's Syndrome, Cholelithiasis, Gallstone Ileus, Gastric Outlet Obstruction, Intestinal Obstruction

Introduction

Gallstone ileus accounts for 1 to 4 percent of all cases of intestinal mechanical obstruction. Usually, gallstones pass into the small bowel through a bilioenteric fistula, as a complication of recurrent episodes of cholecystitis¹⁻⁴.

The capacity of a gallstone to cause an intestinal obstruction depends on its size. Gallstones less than 2,5 centimeters in diameter have low risk of causing an intestinal obstruction and are expelled with stool, whereas larger gallstones have a higher chance to impact either at the level of the ileo-cecal valve or in the jejunum¹⁻⁴. Just in less than 10 % of cases of gallstone ileus, a gallstone may remain stuck in the duodenum due to its remarkable size¹⁻⁴.

This rare condition of gastric out-let obstruction is called Bouveret's syndrome.

In the present review the Authors describe two challenging cases of Bouveret's syndrome. Both patients were treated in our Institution. According to patient's

Pervenuto in Redazione Luglio 2017. Accettato per la pubblicazione Settembre 2017:

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advanced ages and severe comorbidities we decided upon endoscopic treatment. The intent was to minimize complications and to avoid surgery, however, the endoscopic approach was unsuccessful in both cases and patients were referred to surgery.

The challenging endoscopic and clinical management prompt the current study.

We review all the published cases of Bouveret's syndrome between 2006, year of publication of the largest case series ⁵, and 2015 with particular regard to the operative management and treatment options.

Materials and Methods

Electronic search was performed using Medline, Embase and PubMed with the following key words: Bouveret's syndrome, gallstone ileus, gastric outlet obstruction, intestinal obstruction. All case reports and case series published in English between 2006 and 2015 were taken in account with particular regard on treatment strategies. Data are presented in Table I.

CASE N. 1

A 88 year old woman was admitted for acute epigastric pain and vomiting. Her past medical history was remarkable for a stroke one year before. She was on antiplatelets agents (dipyridamole and aspirin). On admission, ECG and troponin level were normal and acute cardiological issues were ruled out. On examination abdomen was soft and mildly distended in the upper quadrants with no tenderness or peritonism. Bloods tests were within normal range except for haemoglobin that was 10.9 gr/dl. On abdominal ultrasound scan, air in the biliary tree

was seen whereas the gallbladder was not detectable. A contrast computed tomography confirmed pneumobilia in the left branches of intrahepatic biliary tree and demonstrated a fistula between the fundus of the gallbladder and the second portion of the duodenum. The presence of bowel dilatation above the duodenum was suspicious of proximal biliary ileus.

A decompressive nasogastric tube was inserted and gastrografin swallow was performed; the fistula was clearly visualised and complete duodenal obstruction due to a gallstone was demonstrated (Fig. 1). A try with gastro-duodenoscopy was made to retrieve the stone; however, the endoscopic approach was ineffective. The patient underwent surgical removal of the stone (Fig. 2) via duodenotomy; duodenum was repaired by direct suture and the gallbladder was left untouched. Postoperative course was regular except for a wound infection; the patient was discharged on 14th postoperative day.

CASE N. 2

A 83 year old woman complaining of 'coffee ground' vomiting and dark stools ongoing for 5 days presented to our attention. Her past medical history was remarkable for previous episodes of gastritis and chronic atrial fibrillation on warfarin treatment. On admission an INR of 4.6 was found; hemoglobin was 9.4 gr/dl. Clinical examination revealed no abnormalities. A nasogastric tube was inserted for repeated vomiting and a plain abdominal X-ray showed dilated stomach and a nodular calcific opacity in the right upper quadrant suggestive for proximal biliary ileus (Fig. 3). At this stage, a gastro-duodenoscopy was performed after warfarin had been suspended and INR normalized. The investigation confirmed an impacted gallstone in the duodenum (Fig. 4) and a cholecystoduodenal fistula; no source of bleeding

TABLE I - First line treatment modalities of 49 cases of Bouveret's syndrome published from 2006 until 2015.

	Total number	N. success	N. succ./Total n.
Case reports	49		
Spontaneous resolution	/		
Extra-corporeal shock wave lithotripsy (ESWL)	/		
Surgery ¹²⁻²² 17			
Simple endoscopic stone retrieval ^{6,23-32,21}	13	1	8%
Singular endoscopic approach ^{33-43,7,44}			
Mechanical lithotripsy	10	6	60%
Electro-hydraulic lithotripsy	3	2	67%
Laser lithotripsy	1	/	0%
Multi-modal approach ⁸⁻¹²			
Electro-hydraulic + mechanical lithotripsy	1	1	100%
Electro-hydraulic + laser lithotripsy	1	1	100%
Laser lithotripsy + ESWL	1	1	100%
Laser + mechanical lithotripsy+ ESWL	1	1	100%
Laser + electro-hydraulic + mechanical lithotripsy+ ESWL	1	/	0%



Fig. 1: Upper gastrointestinal tract series on admission. Upper gastrointestinal tract radiography after contrast material oral intake shows a large stone stuck in the duodenum with a fistula between the fundus of the gallbladder and the duodenum (arrow).

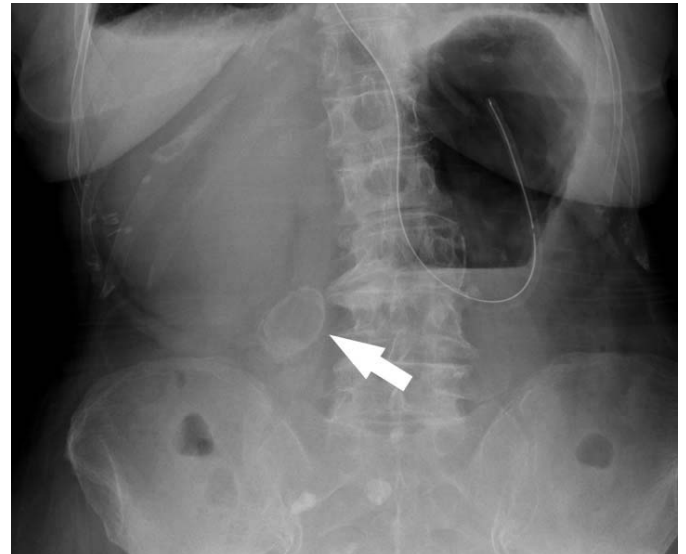


Fig. 3: Plain abdominal X-ray on admission. Abdominal plain film shows dilated stomach and a nodular calcified opacity in the right upper quadrant (arrow).

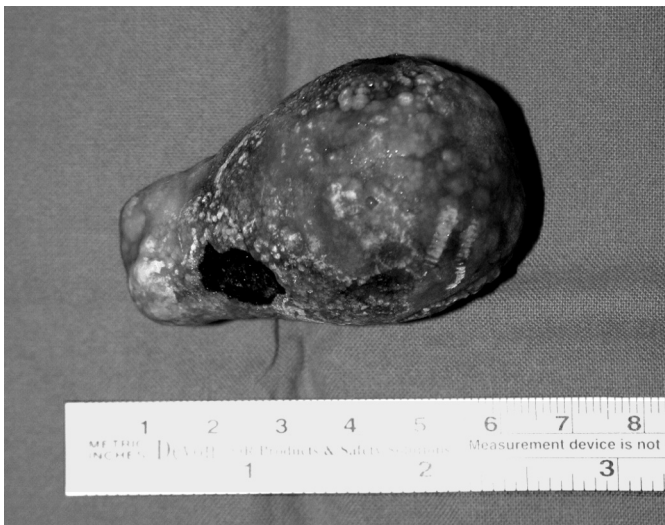


Fig. 2: Surgery. Gallstone after surgical removal via duodenotomy.

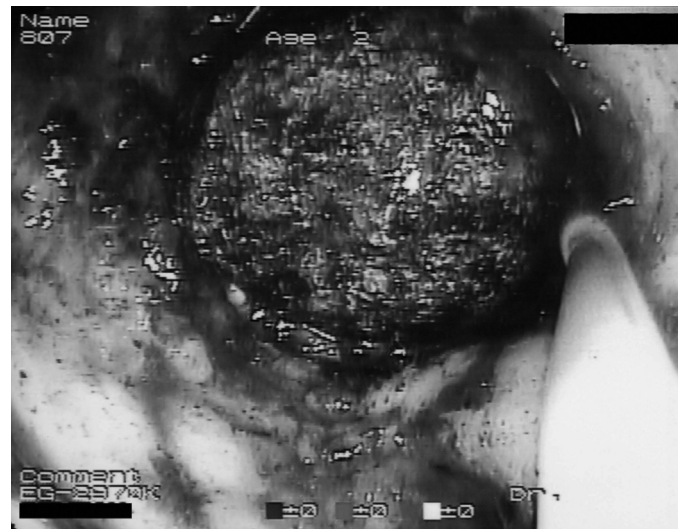


Fig. 4: Gastrointestinal endoscopy. Endoscopic retrieval procedure shows a large gallstone obstructing the duodenal lumen.

was identified. Endoscopic fragmentation was attempted with a mouse tooth forceps unsuccessfully and the stone slipped over the third duodenal portion. Although the gallstone kept progressing in the duodenum and small bowel, as demonstrated by subsequent gastrografin swallow, the patient showed no signs of clinical improvement and surgery was planned. On laparotomy, the stone was found 70 centimeters below the

Treitz ligament and extracted via enterotomy and direct suture. Postoperative recovery was complicated by bleeding at the site of suture that occurred on sixth postoperative day, after low-molecular-weight heparin dose was optimized for atrial fibrillation. A surgical revision with small bowel resection was required. The patient recovered slowly but steady and was discharged 30 days after admission.

Discussion

Clinical presentation in Bouveret's syndrome is nonspecific. Usually symptoms begin one week before medical consultation and a history of cholelithiasis or cholecystitis is often reported. Common symptoms include nausea, vomiting, abdominal pain, anorexia and hematemesis^{4,5}.

Basic imaging workup includes plain abdominal film, upper gastrointestinal series, abdominal ultrasound, esophagogastroduodenoscopy (EGDS) and abdominal computer tomography (CT)^{4,5}.

Operative management of Bouveret's syndrome is challenging; the main goal is to remove the impacted stone with the less invasive procedure.

In the two case reports presented in this article, we treated both patients with an endoscopic approach as first line treatment due to advanced ages and severe comorbidities. The intent was to minimize complications and to avoid surgery, however, the endoscopic approach was unsuccessful in both cases.

Despite gallstones extraction through endoscopy seems at first a reasonable approach, success rate is very low. Cappel MS et al., in their review of 128 cases of Bouveret's syndrome, reported a 10% success rate of endoscopic stone retrieval⁵. In our review of case reports published from 2006 and 2015, we found only one case of successful endoscopic stone retrieval (Table I).

Endoscopic lithotripsy has been reported to be more effective than endoscopic stone retrieval alone. Cappel MS. et al. reported five cases of endoscopic extraction with laser lithotripsy with a 60% success rate (3/5) and twelve cases of endoscopic extraction with mechanical and electrohydraulic lithotripsy with a 25% success rate (3/12)⁵; only one case of extra-corporeal shock wave lithotripsy (ESWL) was described⁵.

According to the reports published from 2006 until 2015 regarding the endoscopic management of Bouveret's syndrome, two different approaches are generally advocated: singular endoscopic approach vs multi-modal approach (Table I).

In the singular approach, patients were treated by fragmentation of the stone using one singular lithotripter in one or more sessions. Success rate for mechanical and electrohydraulic lithotripsy were 60% (6/10) and 67% (2/3) respectively.

Laser lithotripsy was reported in two cases without success and no case of extra-corporeal shock wave lithotripsy (ESWL) has been described as a non-invasive approach.

In case of endoscopic failure, patients were referred to surgery.

In the multi-modal approach, patients were treated in multiple sessions using a combination of different devices and techniques, included ESWL. Out of five cases reported, success rate was 80% (4/5); in one case, the patient developed gallstone ileus and required surgical intervention.

Surgery is considered the definitive treatment of Bouveret's Syndrome⁴⁻⁶. However, no standardized surgical technique has been described yet. If the impacted gallstone is easy to mobilize, the procedures most frequently described include milking of the stone in the jejunum or in the stomach and extraction with simple enterotomy or gastrotomy. Duodenotomy is preferentially avoided for the higher risk of dehiscence unless the stone is firmly impacted in the duodenal lumen. In our first case, the gallstone was entrapped within the duodenal lumen at the level of the aorto-mesenteric compass with no chance to mobilize it and a simple duodenotomy with a direct suture was performed. In our second case, a enterolithotomy was performed after the gallstone had been mobilized following the endoscopic procedure.

In both our cases we left the gallbladder and the fistula colecisto-duodenal intact. There is a general consensus among authors regarding the conservative treatment of cholecysto-duodenal fistula whose risk of complications appears to be low⁷.

Bouveret's syndrome is a rare clinical entity with a challenging operative management, and surgical or endoscopic treatments should be tailored on patient medical conditions and clinical presentation.

Bouveret's syndrome is a rare clinical condition and data reported in medical literature only derive from single case reports and case series. Until now, no clinical evidence has been reached yet. Although the lack of clinical evidence, endoscopic lithotripsy seem to share a good risk-benefit as first line treatment.

The use of ESWL is controversial: despite its non-invasiveness, the risk of worsen patient condition by inducing a gallstone ileus appear reasonable, especially when not accompanied by endoscopy.

Gallstone extraction without fragmentation could result technically challenging, and time-consuming; it could also increase the risk of developing iatrogenic complications such as duodenal perforation and gallstone ileus. Simple endoscopic retrieval should be avoided, especially with gallstones of remarkable size.

If endoscopic approach is ineffective, surgical treatment should be considered.

Riassunto

La sindrome di Bouveret è una rara forma di ileo biliare dovuta ad un calcolo che ostruisce il lume duodenale. Tale condizione clinica si manifesta generalmente nelle persone anziane con una storia anamnestica caratterizzata da plurimi episodi di colecistite acuta. Il calcolo solitamente migra nel lume duodenale attraverso una fistola colecisto-duodenale. La fistola fra la colecisti e la parete del duodeno si manifesta quale conseguenza di una lesione da decubito sostenuta dalle considerevoli dimensioni del calcolo e dai ripetuti episodi di infiammazione.

Gli Autori descrivono due casi di sindrome di Bouveret accompagnati da una revisione della letteratura medica ponendo particolare attenzione agli aspetti operativi.

A causa della rarità di tale entità clinica, i dati reperibili dalla letteratura derivano unicamente dalla pubblicazione di singoli casi, pertanto non esistono trial clinici randomizzati e precise indicazioni terapeutiche.

Gli Autori condividono l'opinione per cui la litotriassia endoscopica sia da considerare la scelta più indicata come trattamento di prima linea. Al contrario, l'estrazione endoscopica del calcolo senza frammentazione può essere impiegata solo se le dimensioni del calcolo lo permettono.

L'impiego di una litotritore extracorporeo è da valutare solo in casi altamente selezionati in quanto potrebbe aumentare il rischio di complicanze iatrogene. In caso di insuccesso della terapia endoscopica, l'approccio chirurgico è da considerarsi mandatorio.

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