Online Submissions: wjg.wjgnet.com
www.wjgnet.com
wjg@wjgnet.com

World J Gastroenterol 2008 January 7; 14(1): 125-128
World Journal of Gastroenterology ISSN 1007-9327

CASE REPORT

Intermittent gastric outlet obstruction due to a gallstone migrated through a cholecysto-gastric fistula: A new variant of "Bouveret's syndrome"

Dimitry Arioli, Ivo Venturini, Michele Masetti, Elisa Romagnoli, Antonella Scarcelli, Pietro Ballesini, Athos Borghi, Alessandro Barberini, Vincenzo Spina, Mario De Santis, Fabrizio Di Benedetto, Giorgio Enrico Gerunda, Maria Luisa Zeneroli

Dimitry Arioli, Ivo Venturini, Elisa Romagnoli, Pietro Ballesini, Athos Borghi, Maria Luisa Zeneroli, Medicina Interna Ⅱ, Department of Medicine and Medical Specialties, Azienda Ospedaliera-Policlinico, Università di Modena e Reggio Emilia, Modena 41100, Italy
Michele Masetti, Fabrizio Di Benedetto, Giorgio Enrico Gerunda, Liver and Multivisceral Transplant Center, Azienda Ospedaliera-Policlinico, Università di Modena e Reggio Emilia, Modena 41100, Italy
Antonella Scarcelli, Gastrointestinal Unit, Department of Medicine and Medical Specialties, Azienda Ospedaliera-Policlinico, Università di Modena e Reggio Emilia, Modena 41100, Italy
Alessandro Barberini, Vincenzo Spina, Mario De Santis, Department of Radiology and Liver and Multivisceral Transplant Center; Azienda Ospedaliera-Policlinico, Università di Modena e Reggio Emilia, Modena 41100, Italy
Correspondence to: Dr. Ivo Venturini, Medicina Interna Ⅱ, Dipartimento di Medicine e Specialità Mediche, Università di Modena e Reggio Emilia, Via del pozzo 71, Modena 41100, Italy. ivo.adsl@alice.it
Telephone: +39-59-4222237 Fax: +39-59-4222360
Received: September 3, 2007 Revised: October 15, 2007

Abstract

Bouveret's syndrome, defined as gastric outlet obstruction due to a large gallstone, is still one of the most dramatic biliary gallstone complications. Although new radiological and endoscopic techniques have made pre-surgical diagnosis possible in most cases and the death rate has dropped dramatically, "one-stage surgery" (biliary surgery carried out at the same time as the removal of the gut obstruction) should be still considered as the gold standard for the treatment of gallstone ileus. In this case, partial gastric outlet obstruction resulted in an atypical and insidious clinical presentation that allowed us to perform the conventional one-stage laparatomic procedure that completely solved the problem, thus avoiding any further complications.

Key words: Bouveret's Syndrome; Biliary gallstone; Gastric outlet obstruction; Biliary surgery

http://dx.doi.org/10.3748/wjg.14.125

Arioli D, Venturini I, Masetti M, Romagnoli E, Scarcelli A, Ballesini P, Borghi A, Barberini A, Spina V, De Santis M, Di Benedetto F, Gerunda GE, Zeneroli ML. Intermittent gastric outlet obstruction due to a gallstone migrated through a cholecysto-gastric fistula: A new variant of "Bouveret's syndrome". *World J Gastroenterol* 2008; 14(1): 125-128

http://www.wjgnet.com/1007-9327/14/125.asp

INTRODUCTION

Gallstones are completely asymptomatic in the majority of patients (60%-80%)[1]. When they become symptomatic, biliary colic is usually the first manifestation to be encountered. Patients with mild symptoms have a higher risk of developing both common and less frequent complications[2].

Biliary fistula is a rare complication (3%-5%) that is frequently preceded by an episode of acute cholecystitis[3,4]. Biliary fistula is mostly encountered in the duodenum although it can occur anywhere in the gastrointestinal (GI) tract[5]. Finally, just 7%-10% of biliary fistulae cause gallstone ileus, an intestinal obstruction caused by a stone that has migrated through the fistula and stopped anywhere in the GI tract[6]. The terminal ileum and ileocecal valve are the most common locations[7], whereas gastric outlet obstruction from an impacted gallstone named "Bouveret's syndrome", is a very rare complication (1/10 000 cholelithisiasis)[8].

To our knowledge, this is the first case of incomplete gastric outlet obstruction manifested as a "waxing and waning" syndrome for a period of over 20 mo before diagnosis was defined.

CASE REPORT

A 79-year old woman was referred to our hospital when a recent oesophagogastroduodenoscopy (OGDS) revealed the presence of a foreign body with its maximum diameter > 4 cm, partially obstructing the gastric lumen, firmly attached to the antral greater curvature region where a fistula was partially explorative. The procedure was performed to investigate the patient's two-year old dyspepsia. This mild symptom alternated every two months with bouts of colic pain associated with self-

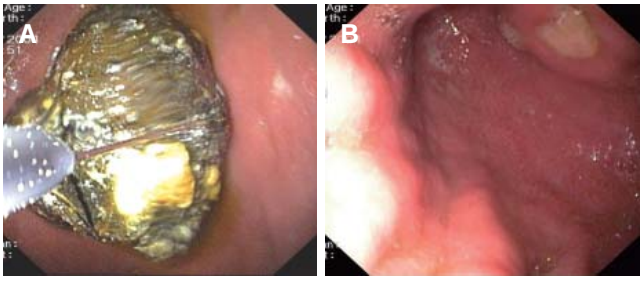

Figure 1 Oesophagogastroduodenoscopy confirming the presence of a foreign body looking like a biliary stone (**A**) and a 15 mm-wide ulcer (**B**).

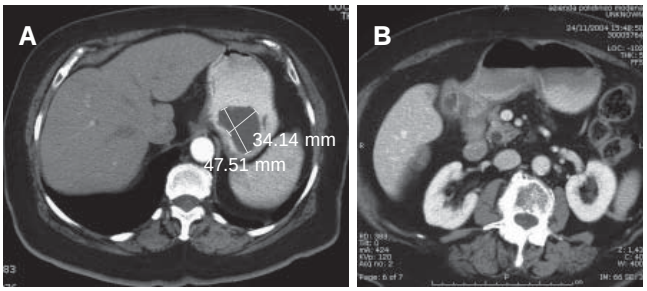

Figure 2 Computerised tomography revealing the density of foreign body typical of ectopic biliary stone (**A**) and the oedematous wall of the gastric antrum (**B**).

limited vomiting lasting 2 d.

The patient reported an earlier negative colonoscopy, performed due to the migration of gastric foreign body through a gastro-colic fistula. His symptoms occurred after two days of acute abdominal pain associated with vomiting due to acute gastroenteritis. An abdominal ultrasound scan (US) revealed cholelithiasis.

The patient was admitted to our ward in good general clinical conditions. Blood tests were completely normal apart from ferrittin, which was twice the normal range.

An abdominal computerised tomography scan (CT) and another OGDS were then performed. Endoscopic examination confirmed the presence of the foreign body, looking like a biliary stone (Figure 1A). Simple endoscopic lithotomy was not possible as the stone was too large, and an attempt at mechanical fragmentation of the stone was unsuccessful. The foreign body appeared to be firmly attached to the gastric wall, and a 15 mm wide ulcer was visible where the opening of the fistula has been previously described (Figure 1B). The CT revealed that the foreign body had a density typical of an ectopic biliary stone (Figure 2A) and the oedematous wall of the gastric antrum could not be separated from the thickened wall of the gallbladder fundus (Figure 2B).

Further air-fluid level was present in the gallbladder (Balthazar and Schester sign) and the intra- and extrahepatic bile ducts were slightly stretched. Lastly, a magnetic resonance cholangio-pancreatography (MRCP) was performed in the hope of observing the fistula. The procedure was negative (Figure 3). All of these data suggest that a multidisciplinary consultation was needed to decide the most appropriate management for this particular clinical setting.

We eventually opted to attempt "one-stage surgery".

Figure 3 Magnetic resonance cholangiopancreato-graphy showing no fistula.

Figure 4 One-stage surgery with dissection of omental-cholecystic adhesions.

Figure 5 A large ectopic stone extracted.

After dissection of many omental-cholecystic adhesions, retrograde cholecystectomy was performed successfully (Figure 4). Finally, a gastrostomy was performed on the anterior gastric wall and a large ectopic stone was extracted (Figure 5).

The post-surgical course was regular and during six-month follow-up the patients' general condition was excellent. The patient was completely asymptomatic with normal biliary tree on the US scan.

DISCUSSION

Like any other gallstone ileus, "Bouveret's syndrome" defined as gastric outlet obstruction caused by a large gallstone occluding the pyloro-duodenal region following a biliogastric or bilioduodenal fistula, primarily affects the geriatric population, with a medium age of 65-75 years and a female predominance (3-16:1)[9-11]. For these reasons, up to 80%-90% of patients have concomitant medical

illnesses (CIC, IRC, diabetes) and the management of such patients should be guided by their general clinical status[4].

The long period of approximately twenty months between the onset and diagnosis of the disease, can be attributed to the patients' carelessness, and the alternating gravity of symptoms combined with the patient's good general health can justify the decision to perform one-stage surgery.

In our case, the particular position of the gallstone led the patient to an insidious clinical setting that urged us to opt for "one-stage surgery" (namely, biliary surgery carried out at the same time as the removal of the intestinal obstruction), which should be still considered as the gold standard for the treatment of gallstone ileus[12]. In fact, it is the only procedure to solve the problem and avoid recurrences and other complications of gallstone disease[13].

In any case, over the past two decades, technological progress has probably made lithotomy or lithotripsy a more conservative approach to reducing patients' risks and has thus been suggested for older patients in poor medical conditions. In our patient, simple endoscopic lithotomy or lithotripsy was not indicated as the stone was excessively large (4 cm) and the inflammatory aspects of the gastric antrum required surgical treatment in order to close the fistula. An attempt at endoscopic mechanical fragmentation of the stone was unsuccessful.

The so-called "two stage surgery" consists of postponing biliary surgery (cholecystectomy and fistula repair) to a later stage with the presence of residual symptomatic stone in asymptomatic subjects[12,24].

Although no patients deny prompt relief from an intestinal obstruction[4], in the case of Bouveret's syndrome, an endoscopic approach represents a reliable alternative to gastro/duodenothomy[15,16]. Preoperative aetiological diagnosis is now possible in most patients and in this case, endoscopic lithotomy and lithotripsy (where possible) represent the first-line approach to treatment as the death rate associated with surgery is still 19%-24%[17,18].

Nevertheless, surgery remains the main procedure in particular situations such as stone impaction in the fistula[19], stone compression of the duodenal wall[20], GI haemorrhage[21] and improper stone manipulation[18,22-25].

In our case, the medical history and endoscopic findings suggested an unusual chronic situation to which the only possible solution was surgery. We, therefore, performed a complete radiological study to better define the anatomy of the lesion before the surgical procedure, including abdominal CT and MRCP of the biliary tree. The first combines the qualities of US and plain X-rays in defining Rigler's diagnostic triad (intestinal obstruction, pneumobilia and ectopic stone)[4,26]. It, therefore, represents the single best imaging technique for the diagnosis of gallstone ileus and Bouveret's syndrome[26]. It may also be helpful in assessing gallbladder wall thickness (signs of acute or chronic cholecystitis), content (air, residual gallstones) and biliary fistula[5,26].

MRCP has recently been proposed as a useful tool for differentiating between fluid and gallstones and also for observing the fistula when sufficient fluid is present[27].

In our case, all the investigations suggested a complicated clinical situation in which an incomplete pyloric occlusion developed following the migration of a large stone through a cholecystogastric fistula, probably due to a poorly-interpreted and treated episode of acute cholecystitis in a case of undiagnosed chronic cholecystitis.

In conclusion, the clinical picture and anatomy of the lesions suggest that a conventional one-stage laparatomic procedure can completely solve the problem, simultaneously avoiding recurrence and other gallstone disease complications.

REFERENCES

1 **Haris HW**. Biliary System. In: Norton JA, Thompson RW, editors. Surgery Basic Science and Clinical Evidence. New York: Springer-Verlag, 2001: 553-584
2 **Ahrendt SA**, Pitt HA. Biliary tract. In: Townsend CM Jr, Editor-in-Chief. Sabiston Textbook of surgery, The Biological Basis of Modern Surgical Practice. Section X. Abdomen. 16th ed. Philadelphia: W.B. Saunders Company, 2001: 1076-1111
3 **Salah-Eldin AA**, Ibrahim MA, Alapati R, Muslah S, Schubert TT, Schuman BM. The Bouveret syndrome: an unusual cause of hematemesis. *Henry Ford Hosp Med J* 1990; **38**: 52-54
4 **Abou-Saif A**, Al-Kawas FH. Complications of gallstone disease: Mirizzi syndrome, cholecystocholedochal fistula, and gallstone ileus. *Am J Gastroenterol* 2002; **97**: 249-254
5 **Bortoff GA**, Chen MY, Ott DJ, Wolfman NT, Routh WD. Gallbladder stones: imaging and intervention. *Radiographics* 2000; **20**: 751-766
6 **Leopaldi E**, Ambrosiani N, Campanelli G. Pyloric stenosis caused by gallstone (Bouveret's syndrome). Presentation of a further case. *Minerva Chir* 1991; **46**: 405-409
7 **Foss HL**, Summers JD. Intestinal obstruction from allstones. *Ann Surg* 1942; **115**: 721. b2-b735
8 **Wakefield EG**, Vickers PM, Walters W. Cholecystoenteric fistulas. *Surgery* 1939; **5**: 674-677
9 **Bouveret L**. Stenose du pylore adherent a la vesicule. *Rev Med* (Paris) 1896; **16**: 1-16
10 **VanLandingham SB**, Broders CW. Gallstone ileus. *Surg Clin North Am* 1982; **62**: 241-247
11 **Masson JW**, Fraser A, Wolf B, Duncan K, Brunt PW, Sinclair TS. Bouveret's syndrome: gallstone ileus causing gastric outlet obstruction. *Gastrointest Endosc* 1998; **47**: 104-105
12 **Pavlidis TE**, Atmatzidis KS, Papaziogas BT, Papaziogas TB. Management of gallstone ileus. *J Hepatobiliary Pancreat Surg* 2003; **10**: 299-302
13 **Clavien PA**, Richon J, Burgan S, Rohner A. Gallstone ileus. *Br J Surg* 1990; **77**: 737-742
14 **Rodriguez-Sanjuan JC**, Casado F, Fernandez MJ, Morales DJ, Naranjo A. Cholecystectomy and fistula closure versus enterolithotomy alone in gallstone ileus. *Br J Surg* 1997; **84**: 634-637
15 **Langhorst J**, Schumacher B, Deselaers T, Neuhaus H. Successful endoscopic therapy of a gastric outlet obstruction due to a gallstone with intracorporeal laser lithotripsy: a case of Bouveret's syndrome. *Gastrointest Endosc* 2000; **51**: 209-213
16 **Maiss J**, Hochberger J, Hahn EG, Lederer R, Schneider HT, Muehldorfer S. Successful laserlithotripsy in Bouveret's syndrome using a new frequency doubled doublepulse Nd: YAG laser (FREDDY). *Scand J Gastroenterol* 2004; **39**: 791-794
17 **Frattaroli FM**, Reggio D, Guadalaxara A, Illomei G, Lomanto D, Pappalardo G. Bouveret's syndrome: case report and review of the literature. *Hepatogastroenterology* 1997; **44**: 1019-1022
18 **Lowe AS**, Stephenson S, Kay CL, May J. Duodenal obstruction by gallstones (Bouveret's syndrome): a review of the literature. *Endoscopy* 2005; **37**: 82-87
19 **Nyui S**, Osanai H, Masuoka H, Ohba S, Ebata T, Yoshida Y. Gastric outlet syndrome caused by a gallstone: report of a case. *Surg Today* 1998; **28**: 412-415
20 **Godiwala T**, Andry M, Varma DG. Unusual cause of gastric

outlet obstruction: CT findings in Bouveret's syndrome. *South Med J* 1988; **81**: 919-921

21 **Chait MM**, Lerner AG. Bouveret's syndrome presenting as upper gastrointestinal hemorrhage. *Am J Gastroenterol* 1986; **81**: 1199-1201

22 **Dumonceau JM**, Delhaye M, Deviere J, Baize M, Cremer M. Endoscopic treatment of gastric outlet obstruction caused by a gallstone (Bouveret's syndrome) after extracorporeal shock-wave lithotripsy. *Endoscopy* 1997; **29**: 319-321

23 **Farman J**, Goldstein DJ, Sugalski MT, Moazami N, Amory S. Bouveret's syndrome: diagnosis by helical CT scan. *Clin Imaging* 1998; **22**: 240-242

24 **Alsolaiman MM**, Reitz C, Nawras AT, Rodgers JB, Maliakkal BJ. Bouveret's syndrome complicated by distal gallstone ileus after laser lithotropsy using Holmium: YAG laser. *BMC Gastroenterol* 2002; **2**: 15

25 **Gencosmanoglu R**, Inceoglu R, Baysal C, Akansel S, Tozun N. Bouveret's syndrome complicated by a distal gallstone ileus. *World J Gastroenterol* 2003; **9**: 2873-2875

26 **Lassandro F**, Gagliardi N, Scuderi M, Pinto A, Gatta G, Mazzeo R. Gallstone ileus analysis of radiological findings in 27 patients. *Eur J Radiol* 2004; **50**: 23-29

27 **Pickhardt PJ**, Friedland JA, Hruza DS, Fisher AJ. Case report. CT, MR cholangiopancreatography, and endoscopy findings in Bouveret's syndrome. *AJR Am J Roentgenol* 2003; **180**: 1033-1035

S- Editor Liu Y **L- Editor** Wang XL **E- Editor** Liu Y