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### Bouverets syndrome: Its characteristics – A review

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#### A B S T R A C T

Bouveret's syndrome is a rare variant of gallstone ileus and a rare cause for the gastric outlet obstruction secondary to an impacted stone in the duodenum. Its presentation is vague and clinical diagnosis is often difficult resulting in a delay in diagnosis. It causes significant morbidity and mortality especially in elderly patients. A spectrum of diagnostic modalities must be made use of in diagnosing this disease. Diagnostic and treatment strategies must be individualised for optimal management and outcome. The surgical approach includes either enterolithotomy alone or in combination with simultaneous or subsequent cholecystectomy and fistula repair. This paper caters the need of characteristics of this disease and a surgical strategy for optimized individual treatment based on the available literature.

### Introduction

Bouveret's syndrome is a rare variant of gallstone ileus (Koulaouzidis and Moschos, 2007; Deepak Joshi et al., 2007), is a fatal condition if not diagnosed early. It was first published by Bouveret's in 1896 (Bouveret, 1896). It is a disease, where gallstone gets impacted in the duodenum causing small bowel obstruction and presents with non-specific upper GI symptoms (Cappell and Davis, 2006). It is most commonly seen in elderly females with a median age of 74 years (Kurtz et al., 1983). The disease presents as subvariants in few cases as

pancreatitis, purulent fistula or severe esophagitis (Rami Bonam et al., 2014; Bhama et al., 2002). CT abdomen clinches the diagnosis in most of the cases with subsequent endoscopic visualization of non-fleshy mass at obstruction site. Endoscopic removal and surgical treatment with enterolithotomy forms the main stay of treatment. An attempt is made to define the characteristics of this disease with early diagnosis and treatment modalities to minimize the morbidity and mortality. A

thorough review of literature and standard reference books has been made.

### **Bouveret's syndrome**

Gall stone disease is one of the common gastrointestinal diseases. Gall stone ileus is a rare complication of cholelithiasis about 0.5% (Liew et al., 2002) Bouveret's syndrome is a form of gallstone ileus that occurs due to impaction of gallstone in stomach or duodenum traversing through the bilio-enteric fistula causing gastric outlet obstruction. This is more prevalent in elderly females with a median age of 74 years and female to male ratio of 1.9 (Cappell and Davis, 2006; Kurtz et al., 1983). Mortality and morbidity has been significant accounting up to 12–15%.

Bilio-enteric fistulas are reported only in 3 to 5% of gallstone diseases (Beuran et al., 2010). Gallstones those pass through the cholecystoenteric fistula i.e., either from gallbladder or bile ducts into the duodenum usually pass distally without any obstruction of the bowel. Chronic cholecystitis is the major underlying cause for the formation of fistula (Giuseppe S. Sica et al., 2005). Most common fistulas observed are cholecystoduodenal (67%) followed by cholecystocolic (17%), cholecystogastric (5%) and choledochoduodenal fistulas (5%) (Clavien et al., 1990). Stones measuring less than 2.5 cms usually pass down spontaneously without any complications. Many of the larger stones cause obstruction at terminal ileum in about 80% cases (Cappell and Davis, 2006), proximal jejunum and ileum and colon in 20% cases and in duodenum in 3% of the patients (Clavien et al., 1990).

The obstruction by the gallstone initiates the chronic inflammation in the walls of both bowel and biliary system. This is followed

by increase in the intraluminal pressure causing local ischemia and necrosis of the walls allowing stones to perforate into the bowel from the biliary system.

### **Presentation**

The presenting symptoms of Bouveret's syndrome are very non-specific and variable. The most common symptoms noted as a clinical triad are epigastric pain, nausea and vomiting (Shalowitz, 1989). The other non-specific symptoms usually seen are abdominal fullness, fever and rarely hematemesis (Cappell and Davis, 2006). On clinical examination, pyrexia, abdominal tenderness, dehydration, abdominal distension is the non-specific signs which are noted. The differential diagnoses that should be kept in mind are pancreatitis, perforated peptic ulcer, bezoars, diverticulae, fibrotic ulcers and neoplasia. Few cases have been reported where the subject presents with pancreatitis with high serum amylase and lipase levels. Possible explanation for this presentation would be obstruction of pancreatic duct exit into duodenum by the gallstone (Liew et al., 2002). Few cases present as subvariants as purulent fistula, esophagitis or in sepsis and massive arterial bleed from eroded cystic artery.

### **Diagnostic approach**

Appropriate diagnostic modalities that confirm the diagnosis ruling out other common causes must be sought for with thorough clinical examination (Toth et al., 2013). Chest and abdominal plain radiographs must be taken which show the Rigler's triad comprising of small bowel obstruction, ectopic gallstone and pneumobilia in 30–35% cases (Trubek et al., 2001). Ultrasound abdomen study reveals gastric dilation, acute inflammation of

gallbladder in suspicion of Bouveret's syndrome. The probable disadvantages of this modality are pathology might be obscured because of extensive bowel gas and in locating the ectopic gallstone. CT abdomen is always recommended which throws light on the fistula formation, pneumobilia, inflammatory reactions surrounding the structures and location, size and number of obstructing gallstone CT abdomen is superior to plain radiographs in finding Rigler's triad (Brennan et al., 2004). In 15–18% cases, the stones are isoattenuating, where CT abdomen may not differentiate stone from surrounding bile. Hence, MRCP is recommended in such cases which delineate the isoattenuating stones from bile simultaneously fistula track can be studied.

The next important step is esophago gastroduodenoscopy (OGD) where, in 65–69% cases gallstone can be identified in duodenum obstructing the natural path of bolus. By thorough study bilioenteric fistula, dilated stomach with food contents, ulceration and oedematous walls of duodenum and most importantly a non-fleshy mass is seen at obstruction site can be found.

## **Treatment**

Treatment options in this disease are limited with endoscopy and surgical management. Endoscopic retrieval of stone should always be attempted in the beginning and there are fewer complications associated with it when compared with surgical treatment (Marchall and Hayton, 2004; Bedogni et al., 1985; Doycheva et al., 2009). As this disease is seen more commonly in elderly age group, the complications and comorbidities limit further surgical management. The stone size too plays a role; size more than 2.5 cms is difficult to retrieve endoscopically and may

get impacted in oesophagus (Lowe et al., 2005). This is the major reason for failure of endoscopic removal (Cappell and Davis, 2006). Fragmentation of the stone too may cause the distal gallstone ileus (Moschos et al., 2005). Many endoscopic techniques have been tried with varied success rates depending on the cases including mechanical lithotripsy, netextraction, electrohydraulic lithotripsy (Zielinski et al., 2010), intracorporeal laser lithotripsy (Doycheva et al., 2009), or combinations of these techniques (Doycheva et al., 2009). Though these time consuming techniques are more successful in proximal obstruction overall success rate is very less about 9% (Lowe et al., 2005).

The main treatment modality is surgical. When the endoscopic methods fail, the stones are too large and stone has fragmented into smaller pieces causing distal obstruction forms the main indication for surgical treatment (Hussain et al., 2013). Most commonly performed procedures are entero lithotomy and gastrostomy (Cappell and Davis, 2006). It is advantageous to take antimesenteric longitudinal incision with a transverse closure of the same to avoid post-operative stenosis. This can be done alone or combined with cholecystectomy with fistula repair as one stage procedure or subsequently in another setting. Many surgeons prefer not to operate on fistula as the chances of spontaneous closure are high when the bile ducts patent and no stones are noted in gallbladder (Koulaouzidis and Moschos, 2007). The decision to operate on the patient with minimal invasive surgery is totally subjective depending on the individual case and experience of surgeon (Balakrishnan et al., 2008) but no data present to affirm the best approach. In old patients with multiple comorbidities, we advocate one stage procedure in acceptable conditions to avoid post operative

complications. In young patients, late postoperative complications would be high due to minimal co morbid conditions and long expectancy of life. Hence, in optimized patients decision to operate in one stage should be done in absence acute inflammatory conditions. In case of absence of spontaneous closure of fistula, its repair in subsequent setting may cause debilitating post operative biliary symptoms increasing the morbidity and mortality of the patients. Due to advances in diagnostic modalities and knowledge on this disease, mortality has dropped down from 30% to 12–15% (Koulaouzidis and Moschos, 2007). Overall mortality rate does not depend on the duration of surgery but definitely in delay in diagnosis of this disease (Liew et al., 2002).

## Conclusion

Bouveret's syndrome is a variant of gallstone ileus which should be considered in elderly ladies with non-specific symptoms of upper GI with or without past history of gallstone disease. Due to new diagnostic modalities and techniques the diagnosis of this disease is fairly easy and it should be one among the differential diagnoses. Rigler's triad should always arouse the suspicion of Bouveret's syndrome. Endoscopic removal should always be considered initially when indicated. Laparotomy or laparoscopic intervention is the main stay treatment. It is a rare and life threatening condition hence unnecessary investigations and delay in appropriate surgical procedure should be avoided to minimize the morbidity and mortality.

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