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Successful Treatment of Bouveret Syndrome by **Electrohydraulic Lithotripsy and Double Balloon** Endoscopy

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Conflict of interest:		None declared	
Р	atient:	Female, 84	
Final Diagnosis: Symptoms:		Bouveret syndrome Abdominal and/or epigastric pain vomitting	
Clinical Procedure:		Electrohydraulic lithotripsy	
Specialty:		Gastroenterology and Hepatology	
Objective:		Rare disease	
Background:		Bouveret syndrome is a rare complication of cholelithiasis that often leads to symptoms of gastric outlet obstruction.	
Case Report:		An 84-year-old woman developed acute abdominal symptoms due to impaction of a gallstone in the horizon-	
		tal part of the duodenum. The diagnosis was support	ted by abdominal computed tomography and double bal-
		loon endoscopy. Considering her advanced age and t	the position of the calcified gallstone, we decided to per-
		form electrohydraulic lithotripsy using double balloo	on endoscopy for treatment. Finally, the impacted stone
		was removed with reduced size. She was discharged	home 10 days after admission without recurrence.
Conclusions:		This case illustrates that electrohydraulic lithotripsy u	sing double balloon endoscopy is very effective, especially
		for treatment of Bouveret syndrome caused by gallst	tone impaction in the horizontal part of the duodenum.
MeSH Keywords:		Gallstones • Ileus • Lithotripsy	
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Background

Bouveret syndrome is defined as gastric outlet obstruction resulting from the passage and impaction of a gallstone through a cholecystoduodenal fistula into the duodenum. There are no definitive guidelines for its approach and management. Because most patients are elderly with multiple comorbidities, endoscopic therapies have been described [1]. Various kinds of endoscopic modalities have been used for the treatment of Bouveret syndrome [2]. Electrohydraulic lithotripsy (EHL) using double balloon endoscopy (DBE) is considered very effective, especially for the treatment of Bouveret syndrome caused by gallstone impaction in the horizontal part of the duodenum. Furthermore, DBE facilitates easy visualization and access to the stone. Herein, we report the case of an 84-year-old woman who developed acute abdominal symptoms due to impaction of a calcified gallstone in the horizontal part of the duodenum, which was treated by EHL.

Case Report

An 84-year-old woman was admitted to the emergency room with symptoms of frequent vomiting and acute abdominal pain. In the past, she had occasional epigastric pain that disappeared without any treatment. Her past medical history was significant for duodenitis and a 2.5-cm gallstone in the gallbladder. On physical examination, she had severe epigastric pain. Abdominal computed tomography (CT) demonstrated findings consistent with an impacted ectopic rim-calcified gallstone measuring 2.5 cm in the horizontal part of the duodenum (Figure 1A), a dilated, fluid-filled stomach and duodenum, and a contracted gallbladder with fluid accumulation and irregular wall. For primary treatment, a nasogastric tube was placed to decompress the stomach and duodenum. A contrast study using Gastrografin through the nasogastric tube demonstrated a 2.5-cm translucency in the horizontal part of the duodenum (Figure 1B). There was no defluxion of Gastrografin in the anal side of the gallstone. Given her advanced age, an endoscopic intervention was recommended. Esophagogastroduodenoscopy (EGD) was performed primarily. EGD revealed a fistulous sinus at the posterior wall of the duodenal bulb (Figure 2A). Infusion of Gastrografin through the fistulous sinus showed a contrasted gallbladder, cystic duct, and bile duct (Figure 2B). She was diagnosed with Bouveret syndrome. However, EGD could not show the gallstone because the stone was impacted at the inner horizontal part of the duodenum. After obtaining informed consent, we decided to perform long-type DBE (EI-580BT, Fujifilm Corp., Tokyo, Japan). This approach revealed the presence of a large gallstone obstructing the lumen of the horizontal part of the duodenum, as well as a small gallstone at the oral side (Figure 3). We decided to perform EHL with water immersion to break the impacted gallstone after removing the small gallstone in the stomach. EHL was performed with a 3.0 Fr probe and a LithoTron EL 27 Compact (Waltz Electronik, Hamburg, Germany) (Figure 4). The probe was inserted through the biopsy channel and the channel was filled with normal saline solution. By using DBE, we were able to fill the duodenal loop with saline solution to get effective spark discharge. The EHL was a long procedure because much time was needed to break the impacted gallstone, which was composed of an extremely hard inner core and soft outer shell. Repeated shock



Figure 1. (A) Impacted ectopic rim-calcified gallstone measuring 2.5 cm in the horizontal part of the duodenum. (B) Gastrografin through the nasogastric tube demonstrated a 2.5-cm translucency in the horizontal part of the duodenum.



Figure 2. (A) Fistulous sinus at the posterior wall of the duodenal bulb. (B) Infusion of Gastrografin through the fistulous sinus.



Figure 3. Impacted gallstone obstructing the lumen of the horizontal part of the duodenum, as well as a small gallstone at the oral side.

applications gradually fragmented the stone shell. As the EHL probe was exhausted, we used grasping forceps. Finally, the impacted stone with reduced size was removed. Abdominal CT immediately after EHL showed the gallstone moving into the jejunum, and an abdominal CT 2 days after EHL showed that the gallstone had disappeared from the intestine. She was discharged home 10 days after admission, without recurrence. EGD at 5 months after admission showed that the fistulous sinus was healing gradually with deformation of the duodenum,



Figure 4. EHL with water immersion.

and the cholecystoduodenal fistula was judged to be closed. The patient was followed with laboratory and imaging tests.

Discussion

We reported a successful EHL using DBE for treatment of Bouveret syndrome due to the gallstone being located in the horizontal part of the duodenum. Bouveret syndrome is defined as a gastric outlet obstruction resulting from a gallstone impaction due to a fistula between the gallbladder and either the duodenum or stomach. Leon Bouveret reported the first 2 cases of this rare form of gallstone ileus in 1896 [3]. Gallstone ileus is relatively rare, complicating only 0.3%–0.5% of patients with cholelithiasis [3]. The majority of the impacted gallstones are located in the terminal ileum (60–75%), jejunum (20–40%), stomach (14.2%), and duodenum (3.5%) [4]. Bouveret syndrome is extremely rare, accounting for only 1–3% of all cases of gallstone ileus [5]. More than 70% of such presentations occur in patients over 70 years old [1], and these patients usually have many comorbidities [1]. The reported mortality rate of gallstone ileus ranges from 7% to 30% [1]. This high mortality is attributed to factors such as old age or frail condition, multiple comorbidities, delayed diagnosis, and complications after surgery [1].

Rapid and accurate management is needed to diagnose Bouveret syndrome. Abdominal CT is considered superior to plain abdominal radiography or abdominal ultrasonography to diagnose the gallstone ileus, with a sensitivity of up to 93% [6]. In our case, abdominal CT revealing the impacted gallstone in the duodenum enabled us to provide rapid and accurate diagnosis and successful management.

Factors that favor biliary-enteric fistula formation are gallstone size (2–8 cm), long history of biliary disease, recurrence of cholecystitis, female sex, and old age (>60 years) [3]. When the pressure in the gallbladder increases suddenly due to cholecystitis with the impaction of gallstones, the gallbladder adheres to the adjacent structures. Adherence to the necrosis of the gallbladder wall leads to biliary-enteric fistula formation; of these fistulas, 60% are cholecystoduodenal, 17% are cholecystocolic, 5% are cholecystogastric, and 5% are choledochoduodenal [7].

In our case, the patient was diagnosed with a cholecystoduodenal fistula. She had experienced frequent stomachache and had been diagnosed with duodenitis and cholecystolithiasis 2 years earlier. We suggest that chronic cholecystitis with adherence of the gallbladder wall to the duodenum can lead to duodenitis and cholecystoduodenal fistula.

Currently, no definitive guidelines have been proposed to manage Bouveret syndrome. Surgical operations are thought to be the most common treatment. However, a previous study revealed the high risks and complications of the surgical operation [1]. Therefore, endoscopic retrieval of the obstructing gallstone is an appealing option in a high-risk population of elderly patients with comorbidities. In our case, we preferred endoscopic options over surgical operation because of the patient's advanced age. Various endoscopic modalities have been used for the treatment of Bouveret syndrome, including EHL, endoscopic nets/baskets, laser, mechanical, and extracorporeal shockwave lithotripsy [2]. In our case, abdominal CT showed a rim-calcified gallstone. Moreover, breaking a rim-calcified or X-ray positive gallstone is considered difficult because of the rigid calcification. EHL allows breaking large, calcified stones that cannot be treated by other endoscopic modalities [2]. The principle of EHL is the occurrence of an electric high-voltage spark between 2 isolated electrodes located at the tip of a fiber [8]. The electric sparks are delivered in short pulses that create an immediate expansion of the surrounding liquid, including a spherical shock wave [8]. During EHL, the stone should be continuously immersed in water. Bourke et al. reported that the most important problem for effective spark discharge is difficulty in filling the lumen of the duodenum with saline solution using endoscopy and colonoscopy [9]. In our case, inflating the balloon enabled us to easily pool the saline and perform EHL effectively. Furthermore, DBE enabled clear visualization and access to the gallstone, which could not be visualized by EGD. Using DBE, we can perform EHL even if the gallstone passes into the distal small bowel.

To date, there have been some case reports of Bouveret syndrome, but it appears that Bouveret syndrome due to a gallstone in the horizontal part of the duodenum is rare. Dumonceau et al. [10] reported 2 cases of stone impaction in the horizontal part of the duodenum among 61 cases. Fujita et al. [11] reported a case of gallstone ileus due to a gallstone in the horizontal part of the duodenum; they performed duodenoscopy and endoscopic retrograde cholangiopancreatography to observe the gallstone and fistula. In our case, DBE facilitated easy visualization and access to the stone. DBE enabled observation and treatment simultaneously. EHL using DBE is very effective, especially for treatment of Bouveret syndrome caused by a gallstone located in the horizontal part of the duodenum. Use of EHL can avoid postoperative complications, but there is the possibility of distal impaction of gallstone fragments. In our case, abdominal CT immediately after and 2 days after EHL showed that recurrent gallstone ileus had not occurred.

Potential long-term complications of biliary-enteric fistula include biliary infection and gallbladder cancer. Bossart et al. found a 15% incidence of gallbladder cancer in 57 patients undergoing surgery for fistulas [12]. The digestive reflux back into the gallbladder can lead to intestinal metaplasia and gallbladder cancer. Therefore, cholecystectomy and fistula repair are recommended to prevent future malignant transformation [13]. However, studies have shown that fistula repair may be unnecessary, such as in our case, as these fistulas can close spontaneously if the cystic duct is patent and residual gallstones are not present [13]. In our case, because the fistula closed spontaneously and the gallstone was successfully removed, cholecystectomy and fistula repair were not performed, but clinicians must be aware of possible recurrent cholecystitis and risk of gallbladder cancer.

Conclusions

Bouveret syndrome requires a high degree of suspicion for diagnosis in patients reporting symptoms of gastric outlet obstruction. Early diagnosis using abdominal CT is important because of the high mortality rate, mainly in elderly patients with various comorbidities. EHL is a less invasive and useful treatment, especially in cases similar to the presented case of Bouveret syndrome due to a gallstone in the horizontal part of the duodenum. As EHL using DBE provides easy visualization and access to the stone and pooling of the saline. It is a better

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treatment option for patients with Bouveret syndrome due to gallstone impaction in the horizontal part of the duodenum.

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Conflict of interests

None.

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