The Oldest Patient with Gallstone Ileus: Report of a Case and Review of 176 Cases in Japan

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Summary: We report a 91-year-old woman presenting bowel obstruction due to impacted gallstone, who was the oldest patient which has been reported in Japanese scientific literature. The patient was referred to our hospital due to vomiting and abdominal pain. Computed tomography and abdominal X-ray showed dilated loops of small intestine associated with air-fluid levels, pneumobilia, and a calcified mass in the left iliac fossa. After the diagnosis of bowel obstruction due to gallstone was made, an enterotomy and lithotomy was performed under spinal anesthesia. The postoperative recovery was uneventful. We also reviewed 176 cases of gallstone ileus which were reported in the Japanese literature in the past 20 years. The retrospective analysis demonstrated that one-stage enterolithotomy alone may be acceptable as the first choice of operative treatment. The gallstone ileus is a rare, but important disease because urgent and appropriate surgical therapy is required.

Key words gallstone ileus, bowel obstruction, pneumobilia, cholecystoduodenal fistula, surgery

INTRODUCTION

Gallstone ileus is a rare mechanical intestinal obstruction caused by the passage of gallstones into the gastrointestinal lumen. Gallstone ileus accounts for only about 1-3% of cases of mechanical obstruction of the small bowel, but it is a quite important disease especially in elderly patients, as this disease shares 25% of all small bowel obstructions in patients older than 65 years [1]. As the majority of the elderly patients frequently have concomitant diseases and show relatively poor clinical manifestations, it is important to recognize this disease to avoid delayed diagnosis. Furthermore, its high incidence among elderly people may explain its association with chronic and degenerative factors, which may increase the complexity of the treatment choice.

Herein, we present a case of gallstone ileus in a 91-year-old woman with small bowel obstruction, who was successfully treated by surgery. Also, we reviewed the literature written in Japanese for Japanese Medical Journals or abstracts presented at Japanese conferences with available information between 1985 and 2005 using the website of IGAKUCHUO ZASSHI, and analyzing the clinical data for 176 Japanese patients of gallstone ileus. This article may assist the physician and surgeons to further understand this disease, as well as to illustrate diagnostic and therapeutic pitfalls, and determine the current therapeutic strategy based on a detailed analysis of a number of Japanese cases. Notably, our patient was the oldest patient which has been reported in Japanese scientific sources.

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Abbreviation: CT, computed tomography.

CASE REPORT

A 91-year-old Japanese woman was referred to our hospital due to vomiting and abdominal pain. She appeared dehydrated with a fever of 38.3 degrees. Past medical history was unremarkable with no episodes of cholecystitis. No other concomitant disease was determined to be present. Blood pressure was 116/65 mmHg and pulse rate was 74 beats/min. Her abdomen was slightly distended but soft with central and left-sided tenderness without muscle guarding. Bowel sounds were weak but audible. Neither jaundice nor anemia was recognized. Significant laboratory results were: white blood cell count, 10400/ μ L, C-reactive protein, 1.26 mg/mL; total bilirubin, 2.8 mg/dL; lactate dehydrogenase (LDH), 486 IU/L. Plain abdominal X-ray showed marked small bowel distention with air-fluid level, and the presence of a round calcified component in the pelvic cavity (Fig. 1). Computed tomography (CT) of the abdomen revealed pneumobilia and distention of the jejunum, with a high density mass of 2.5 cm in diameter in the alimentary tract (Fig. 2). Air in the gallbladder and an adhesion between the thickened gallbladder wall and duodenal wall was visualized. The targeted lesion was located inside the ileum and consisted of two layers; rim calcification and an internal area of isodensity. There was fluid pooling in the proximal intestine of the high density mass, suggesting the cause of the mass to be bowel obstruction. Based on these radiological and clinical assessments, the diagnosis of bowel obstruction due to gallstone was made. Since transnasal drainage of gastrointestinal tract for decompression was not efficient, the general status of the patient prompted us to undertake a surgical intervention. After obtaining an adequate informed consent, enterotomy and surgical lithotripsy were performed under spinal anesthesia 48 hrs after disease onset. The location of stone was detected by palpation and found to be impacted in the ileum at 50 cm proximal to the terminal ileum. After the stone was removed, the ileum was primarily closed with double-layer running stiches of a bioabsorbable material. The stone was of a combined nature 3 $\times$ 2 cm in size (Fig. 3). The patient's postoperative recovery was uneventful.

DISCUSSION

Since Kasahara et al. [2] reported the retrospective analysis of the clinical features of gallstone ileus in 112 Japanese patients between 1903 and 1978, a single or a few case reports restricted to the Japanese language have been available in Japan. Decades ago,

gallstone ileus was known to be a disease with relatively high mortality rates, however current progress in surgical technique, diagnostic imaging, and innovation of drugs might have improved the outcome of this disease. Therefore, it is important to reevaluate the current feature of the gallstone ileus aiming to clarify the diagnosis and management, and to summarize clinical data from multi-Japanese centers in the English literature.

As far as we know, there have been 176 reported cases of gallstone ileus written in the Japanese literature in these 20 years between 1985 and 2005. The number of male and female patients was 49 and 127, respectively. The age of the patients ranged from 24 to 91 years old. (Fig. 4) Thus elderly people are an important group of patients with gallstone ileus. Typically, the pathogenesis of gallstone ileus is based on acute or chronic cholecystitis associated with cholelithiasis, spreading inflammations and adhesion to alimentary tracts, leading to biliary-enteric fistula. In the United States, about 50% of the patients have a history of gallbladder disease [3]. In our series of Japanese patients, 124 of 176 reported cases (66%) to have the presence of stones in the biliary tracts. Our review of the Japanese literature revealed that cholecystoduodenal fistula is most frequently found in 96.5% of Japanese patients with gallstone ileus. Similarly, cholecysto-duodenal fistulas are those which are most frequently described in the worldwide-reports, followed by cholecysto-colonic, cholecysto-gastric and duodeno-left hepatic duct fistulas [4-6]. In one rare case, the bile duct stone disrupted through the Papilla Vater into the duodenal lumen [7]. Potentially reactive substances in bile juice may react with the intestinal epithelial cells which may induce subsequent impacted stones which are associated with mucosal injuries [8].

Impacted gallstones ranged in size from 2 cm to 10 cm, with a mean of 4.3 cm in the series of Japanese patients. A gallstone is usually more than 2.5 cm in diameter to cause an intestinal obstruction in the normal small intestine [9]. As the smaller stones may be easily evacuated, the impaction of the gallstone smaller than 2 cm is unusual, but, may occur when some pathologic state of the intestine including spasms, angulation or adhesion causes narrowing of the intestinal lumen [10]. The review of Japanese patients revealed that intestinal obstruction by gallstones is seen more frequently at the ileus, followed by jejunum and duodenum (Fig. 5). The impacted stone at the colon is uncommon and occurred in only 2.9% of Japanese patients, which was comparable to that of US reports

Fig. 1. Plain abdominal X-ray showed marked small bowel distention with air-fluid level, and demonstrated a round calcified component in the pelvic cavity.

Fig. 3. The impacted stone was a combined stone 3×2 cm in size.

Fig. 2. Pneumobilia (arrow) and air in the gallbladder are seen (upper panel). CT demonstrated the distention of the jejunum with a high density mass of 2.5 cm in the diameter in the alimentary tract (lower panel).

Fig. 4. Gender and age of the patients with gallstone ileus in Japan.

Fig. 5. The site of the impacted stones of 176 Japanese patients between 1985-2005.

(4.8%) [3]. In elderly patients, there may be concomitant disease such as colonic cancer, resulting in a greater frequency of episodes of disease onset.

Although clinical symptoms are not specific, most of the Japanese patients with gallstone ileus presented with abdominal pain (91.5%), accompanied by vomiting in 59.7% of all patients (Table 1). Jaundice (serum total bilirubin >2.0 mg/dl) was seen in 7.3% of Japanese patients. The use of diagnostic radiological tools has greatly facilitated the clinical diagnosis. The typical radiological images may include pneumobilia, intestinal obstruction, and ectopic gallstone. Pneumobilia may be secondary to bilioenteric fistula, endoscopic sphincterotomy, or an incompetent sphincter of Oddi, therefore it is not a definitive indication for gallstone ileus. Plain abdominal X-ray is a conventional, but useful strategy for detection of air-fluid levels and pneumobilia. Plain abdominal X-ray could visualize air-fluid level in 77.8%, bowel loop dilatation in 88.9%, pneumobilia in 37%, air in gallbladder in 3.7% and ectopic stone in 33% of patients according to the clinical study conducted in Italy [11]. Helical CT may improve the diagnosis of gallstone ileus providing important information [12,13]. Abdominal ultrasound may also prove to be a useful tool for the diagnosis [14,15]. Radiological upper gastrointestinal

tract imaging with barium may show an intraluminal stone or a fistula in the case of a choledocho-duodenal or choledocho-colonic fistula [16]. Our review of the Japanese literature disclosed that air-fluid levels and bowel loop dilatation were most frequently observed in our series [11]. Pneumobilia was observed in 50% of Japanese patients.

The therapeutic management of gallstone ileus remains controversial [17]. However, considering that a spontaneous passage of the impacted stone through the intestine is rare, a surgical approach is likely to be an appropriate therapeutic strategy if the condition of the patients permits such intervention [18]. Enterolithotomy alone was most frequently chosen as the routine surgical treatment for gallstone ileus in Japan. The mortality was higher in the patients with one-stage surgery including enterolithotomy, cholecystectomy and fistula closure, regardless of the patients' age [17]. Therefore simple enterolithotomy is considered to be appropriate in most patients. However, additional performance of a one-stage cholecystectomy and repair of fistula are desirable if the condition of the patient allows it, since there may be a concern that an enterolithotomy alone may cause the complications related to the persistence of the biliary-enteric fistula, which includes the possibility of recurrent of gallstone ileus and recurrent cholangitis [18]. Conservative therapy may also be considered for the treatment of gallstone ileus when the obstructing gallstone is smaller than 2 cm based on radiological measurement. Among 176 Japanese patients, 14.2% (25/176) of patients were conservatively treated. Endoscopic lithotomy was available only when the stone was impacted in the stomach, duodenum or colon. In the present case, we elected to perform enterolithotomy considering the age of the patient and the impacted site of the stone.

The occurrence of a high mortality rate may be due to the frequency of relatively elderly patients with unstable clinical conditions, concomitant disease such as cardiorespiratory disease and/or diabetes mellitus, delayed initiation of the treatment because of uncommon symptoms, and associated with all of these, frequent surgery associated complications such as pneumonia or cardiac failure. Overall mortality of patients was as high as 60% before 1925, 40% in 1960 and 15% after 1970 [3]. Our Japanese literature review indicated that the mortality of this disease has been decreased even further to 8%.

The present case was the oldest patient which has been reported in Japanese scientific literature. Considering the poor conditions due to bowel obstructions and the potential complications related to surgery and

TABLE 1.
Clinical symptoms

Abdominal pain	91.5% (161/176)
Vomiting	59.7% (105/176)
Fever (>38°C)	40.9% (72/176)
Jaundice	7.3% (13/176)
Abdominal distension	84.7% (149/176)

TABLE 2.
Radiological findings

Pneumobilia	50% (88/176)
Air-fluid level	88.1% (155/176)
Detection of stone	31.3% (55/176)

TABLE 3.
Treatments for gallstone ileus

Surgical treatment	
Enterolithotomy	74
Enterolithotomy & fistula closure (one-stage)	44
Enterolithotomy & fistula closure (two-stage)	21
Non-surgical treatment	
Conservative	25
Endoscopic lithotomy	11
Extracorporeal Shock Wave Lithotripsy (ESWL)	1

anesthesia, we performed enterotomy and surgical lithotripsy under spinal anesthesia, which is likely to be most appropriate and minimally invasive for our patient. During approximate 1-year follow up period, the patient has been well without related to the persistence of the biliary-enteric fistula such as cholangitis.

In summary, the successful treatment of the oldest case of gallstone ileus in Japan was reported. Although the clinical symptoms are not specific, it is not difficult to make an accurate diagnosis of gallstone ileus using radiological imaging. The literature review of Japanese patients between 1985 and 2005 has definitively revealed the improved outcome of the disease in the recent years.

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