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Phlegmonous gastritis after biloma drainage: A rare case report and literature review

Phlegmonous gastritis after biloma drainage

Kai-Chun Yang, Hsin- Yu Kuo, Jui-Wen Kang

Abstract

BACKGROUND

Phlegmonous gastritis (PG) is a rare bacterial infection of the gastric submucosa and is related to septicemia, direct gastric mucosal injury, or the direct influence of infection or inflammation on neighboring organs. Here, we present a patient who had spontaneous biloma caused by choledocholithiasis and then PG resulting from bile leakage after biloma drainage.

CASE SUMMARY

A 79-year-old male with a medical history of hypertension had persistent diffuse abdominal pain for 4 d. Physical examination showed stable vital signs, icteric sclera, diffuse abdominal tenderness, and muscle guarding. Laboratory tests showed hyperbilirubinemia and bandemia. Contrast computed tomography (CT) of the abdomen showed a dilated common bile duct and left subphrenic abscess. Left subphrenic abscess drainage revealed bilious fluid, and infected biloma was confirmed. Repeated abdominal CT for persistent epigastralgia after drainage showed gastric wall thickening. The esophagogastroduodenoscopy (EGD) showed edematous, hyperemic gastric mucosa with poor distensibility. The gastric mucosal culture yielded *Enterococcus faecalis*. PG was diagnosed with imaging, EGD findings, and gastric mucosal culture. The patient recovered successfully with antibiotic treatment.

CONCLUSION

PG should be considered in patients with intraabdominal infection, especially from infected organs adjacent to the stomach.

Key Words: Phlegmonous gastritis; Epigastric pain; Choledocholithiasis; Bile leakage; Antibiotics; Case reports

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Core Tip: We report a case of spontaneous biloma caused by choledocholithiasis followed by phlegmonous gastritis (PG) resulting from biloma rupture after biloma drainage. Additionally, we analyzed 44 PG cases reported from 2012 to 2022. The etiology of PG is mainly direct microbial invasion from gastric mucosa injury or hematogenous/Lymphogenous spread and the most important risk factor of PG is an immunocompromised state. In our case, the patient was immunocompetent and PG was caused by bile leakage after biloma drainage rather than the direct influence of infected biloma.

INTRODUCTION

Phlegmonous gastritis (PG) is a rare bacterial infection of the gastric submucosa and is related to septicemia, direct gastric mucosal injury, or direct influence of infection or inflammation in neighboring organs. It is fatal if not diagnosed and treated immediately. An immunocompromised state associated with malignancy, chemotherapy-induced neutropenia, acquired immunodeficiency syndrome, alcoholism, and immunosuppressive drugs are considered the main risk factors^[1,2].

Here, we present a patient with spontaneous biloma caused by choledocholithiasis followed by PG induced by bile leakage after biloma drainage.

CASE PRESENTATION

Chief complaints

A 79-year-old male complained of persistent diffuse abdominal pain for 4 d.

History of present illness

Initially, the abdominal pain occurred abruptly after eating a big meal, 4 d prior to admission. The initial abdominal pain was mainly located in the right upper quarter

abdominal area and then migrated to the whole abdomen. Additionally, the patient experienced nausea, vomiting, constipation, and fever. Recurrent abdominal pain was noted for the 4 d as well. Sonography-guided percutaneous catheter drainage of the left subphrenic abscess, as shown by contrast computed tomography (CT) of the abdomen, was performed. The bilirubin level was 76.0 mg/dL and volume of abscess drainage was around 600 mL. The abscess culture yielded *Enterococcus faecalis* and *Enterobacter cloacae* complex. The blood culture yielded no pathogen isolates. Biloma was confirmed. However, the patient still complained of epigastric pain after drainage.

History of past illness

The patient had a medical history of hypertension and had taken an antihypertensive drug regularly.

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Personal and family history

The patient's personal and family histories were unremarkable.

Physical examination

1 Initial vital signs were a temperature of 37.8 °C, heart rate of 126 beats/min, blood pressure of 163/93 mmHg, and respiratory rate of 32 breaths/min. There was no apparent loss of consciousness. Physical examination showed icteric sclera, abdominal fullness, diffuse tenderness, and muscle guarding. Follow-up vital signs before repeat abdominal CT for persistent abdominal pain 1 were a temperature of 37.5 °C, heart rate of 116 beats/min, blood pressure of 162/84 mmHg, and respiratory rate of 20 breaths/min. Severe muscle guarding and diffuse tenderness were observed.

Laboratory examinations

Abnormal laboratory findings included hyperbilirubinemia (total bilirubin: 3.0 mg/dL, reference range: ≤ 1.2 mg/dL), mildly elevated alkaline phosphatase (138 U/L, reference range: 40-129 U/L), hyponatremia (sodium: 127 mmol/L, reference range:

136-145 mmol/L), impaired renal function (creatinine: 1.59 ⁷ mg/dL, reference range: 0.70-1.20 mg/dL), white blood cell count of 3200/ μ L (reference range: 3400-9500/ μ L), and 8% band form of white blood cells (reference range: 0.0-4.2%).

Imaging examinations

Plain abdominal radiography showed ileus and contrast abdominal CT showed a dilated common bile duct (CBD) and left subphrenic abscess (Figure 1A). Repeat contrast CT of the abdomen was performed for persistent abdominal pain after biloma drainage and showed bile leakage and gastric wall thickening. (Figure 1B). Esophagogastroduodenoscopy (EGD) showed edematous, hyperemic gastric mucosa with poor distensibility (Figure 2A-B). Endoscopic retrograde cholangiopancreatography after EGD showed a dilated CBD with one filling defect of about 10 mm in size (Figure 3A).

FURTHER DIAGNOSTIC WORK-UP

The gastric mucosal culture yielded *Enterococcus faecalis* and the biopsy showed gastric submucosa and mucosa infiltrated by clusters of lymphocytes, neutrophils, and plasma cells (Figure 4A-B). According to initial CT and endoscopic retrograde cholangiopancreatography, the etiology of the initial abdominal pain with fever was a CBD stone with cholangitis and spontaneous biloma. However, according to serial CT images and gastric mucosal culture, the persistent pain after biloma drainage was caused by PG. The etiology of PG was bile leakage after biloma drainage.

FINAL DIAGNOSIS

PG resulting from bile leakage after biloma drainage.

TREATMENT

The patient underwent conservative therapy for PG including parenteral nutrition, biloma drainage, initial broad-spectrum antibiotics (cefepime 2 g, 2 times per day), and

then definitive antibiotics of ampicillin 2 g, 6 times a day for 10 d. Endoscopic retrograde biliary drainage was performed for internal biloma drainage (Figure 3B). Neither endoscopic sphincterotomy nor stone extraction was performed due to coagulopathy.

OUTCOME AND FOLLOW-UP

The abdominal pain was relieved after antibiotic treatment, and follow-up EGD 1 mo later showed normal gastric mucosa with improved distensibility of the stomach (Figure 5A-B). Follow-up abdominal CT 2 mo later showed that the biloma was almost resolved, and the biloma drainage was removed before discharge. The patient is currently being followed up as an outpatient.

DISCUSSION

PG is an infrequent bacterial infection of the gastric submucosa. It was first described in 1862 by Cruveilhier^[3] and an average of one case report per year has appeared over the last 60 years^[4]. We searched for papers indexed in the PubMed database using the keyword “phlegmonous gastritis”. In recent decades, from 2012 to 2022, 44 cases of PG have been reported in the English-language literature, which are summarized in Table 1^[1,4-42]. Our literature review showed that PG affects all age groups (age range: 7-years-old to 89-years-old) and is highly common in the decades of 40s to 70s, with a male-to-female ratio of about 2:1. According to our findings of the literature review, half of the cases (22/44) were in an immunocompromised state, such as alcoholism, diabetes mellitus, acquired immunodeficiency syndrome, chronic hepatitis B, neutropenia after chemotherapy, and treatment with immunosuppressant drugs. Thus, the immunocompromised state appears to be a main risk factor for PG. Other risk factors of PG are prior endoscopic procedure (including for mucosal resection, submucosal dissection, hemostasis, ultrasonography with fine needle aspiration, and mucosal biopsy; 5/44), recent upper airway infection (3/44), malignancy (8/44), and prior gastrectomy or esophagectomy (3/44).

PG type is classified as diffuse or localized, according to the lesion range^[4]. The diffuse type involves the complete stomach, and represents most cases^[43]. In contrast, the localized type is most commonly restricted to the antrum, with rare cases involving the cardia or pylorus. Gastric wall abscess is a localized form of PG^[43]. In our literature view, only 6 Localized type PG cases were identified. The etiology of PG can be classified into primary, secondary, or idiopathic^[17]. Primary PG represents a direct microbial invasion from gastric mucosa injury, which is caused by trauma, malignancy, peptic ulcer, or endoscopic interventions. Secondary PG represents a hematogenous/Lymphogenous spread or direct influence of infection or inflammation in neighboring organs such as infection due to upper airway infection, pancreatitis, or cholecystitis. Idiopathic PG represents an unknown cause with absence of a primary lesion. In our case, the PG was secondary.

The most common clinical presentation of PG is severe and acute epigastric pain accompanied by fever, vomiting, palpitation, melena, and hematemesis. The symptoms of PG mainly occur within 24 h, although they can develop over several days. It is important to differentiate PG from other acute abdomen etiologies such as acute pancreatitis, cholecystitis, and bowel perforation. Diagnostic modalities for PG include EGD, abdominal CT, and endoscopic ultrasonography (EUS). EGD findings show edematous mucosa with fibrinopurulent exudates and superficial ulcerations, loss of rugae, and poor distensibility; however, these features are nonspecific to PG. Several differential diagnoses need to be considered, like acute gastric mucosal lesion, scirrhus gastric cancer, gastric syphilis, corrosive gastritis, malignant lymphoma of the stomach, gastrointestinal stromal tumor, and anisakiasis^[16,35]. Each of these diseases are diagnosed according to the collective findings from EGD imaging examination along with patient data on clinical pattern, medical history and culture test results. CT scan findings include obvious thickening of the gastric wall, and low-intensity areas within the gastric wall^[17]. Of note, EUS has not been routinely recommended but is an excellent tool for detecting and tracking thickening of the gastric wall and degree of

inflammation^[16]. Standard forceps biopsy may not be diagnostic because it does not obtain sufficient submucosal tissue, which is the typically involved layer in PG.

In our review, the most common microorganism was *Streptococcus* spp. (56%, 19/34), followed by *Enterococcus* spp. (10%, 3/34). This result was in line with the 2014 bibliographic review by Veloso-Veloso *et al*^[14].

PG has a ¹ high mortality rate; the key to successful treatment is early diagnosis and therapy. ⁵ Before invention with antibiotics, the mortality rate was 83%-92%^[14]. Kim *et al*^[2] reviewed 36 cases of PG between 1973 and 2003. The mortality rates for surgical intervention and conservative treatment with antibiotics were 20% and 50%, respectively. Recently, Veloso-Veloso *et al*^[14] reviewed 45 cases from 1980 to 2014 and found that the mortality rates for surgically and medically treated patients were 11% and 19%, respectively. In our review of cases reported from 2012 to 2022, the mortality rate was 12% (1/8) for surgery and 8% (3/36) for medical treatment. Through early diagnosis and appropriate antibiotic treatment, the mortality rate of PG has been gradually decreasing but is still high. Surgery should always be considered in refractory cases, which show clinical deterioration despite optimal medical management, and in the presence of complications, such as delayed perforation, abdominal compartment syndrome, bleeding, or stricture^[9,15,17,22,37]. The surgery itself can be a partial or total gastrectomy, according to the range of inflammation^[7]. In our case, conservation therapy with antibiotics alone was successful.

The recurrence rate of PG is low according to our literature review. Only 1 case, which was reported by Taniguchi *et al*^[34], had recurrent PG at 5 d after discharge, and the causes considered most likely were a steroid treatment for allergy and a short-term course of antibiotics. To avoid recurrence, optimizing the process/timing of antibiotic cessation is important and should be determined by laboratory testing data and clinical pattern along with findings from follow-up imaging examinations (*e.g.*, of CT or EUS).

³ A biloma is defined as a collection of bile located outside the bile duct. The main causes of such are iatrogenic or traumatic injuries^[44], with cases of spontaneous biloma being relatively uncommon. The most frequent cause of spontaneous biloma is

choledocholithiasis^[45], with the underlying mechanism hypothesized as an increase in intraductal pressure due to stone obstruction. Unfortunately, the clinical presentations of biloma are non-specific, including abdominal pain, fever, nausea, vomiting, and jaundice^[46].

Nowadays, treatment for spontaneous biloma is nonsurgical, including antibiotics and percutaneous drainage *via* pigtail catheter. ERCP is a feasible alternative with additional benefit because it can not only decompress the biliary tract by endoscopic sphincterotomy and stent placement but also identify the location and severity of an active bile leakage. ³ Surgery is reserved for patients who fail endoscopic stone extraction or present a persistent active leak.

CONCLUSION

This case report describes a rare case of spontaneous biloma caused by choledocholithiasis followed by bile leakage-induced PG after the biloma drainage. PG itself is an uncommon diagnosis of abdominal pain, but should be considered in patients with intraabdominal infection, especially from an infected organ adjacent to the stomach. The key to successful treatment is early diagnosis and initiation of therapy.

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