American Journal of Case Reports

ISSN 1941-5923
© Am J Case Rep, 2017; 18: 125-130
DOI: 10.12659/AJCR.902180

Received: 2016.10.31
Accepted: 2017.01.13
Published: 2017.02.06

A Rare Case of Acute Phlegmonous Esophagogastritis Complicated with Hypopharyngeal Abscess and Esophageal Perforation

Authors' Contribution:
Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
Funds Collection G

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Conflict of interest: None declared

Patient: Female, 60
Final Diagnosis: Acute phlegmonous esophagogastritis complicated with hypopharyngeal abscess • esophageal perforation
Symptoms: Fever • painful swallowing • chest pain
Medication: —
Clinical Procedure: Drainage • debridement • esophageal reconstruction
Specialty: Surgery

Objective: Rare disease

Background: Acute phlegmonous esophagogastritis is a life-threatening disease that may be combined with serious complications. We present the classical radiological and endoscopic features and treatment strategy of a middle-aged female patient suffering from acute phlegmonous esophagogastritis complicated with hypopharyngeal abscess, esophageal perforation, mediastinitis, and empyema.

Case Report: A 60-year-old Taiwanese female presented at our hospital due to fever, fatigue, painful swallowing, and vague chest pain for 5 days. She had a past history of uncontrolled type 2 diabetes mellitus. On physical examination, general weakness, chest pain, odynophagia, and a fever up to 38.9°C were found. Positive laboratory findings included leukocytosis (leukocyte count of 14.58×10^3/μL, neutrophils 76.8%) and serum glucose 348 mg/dL (HbA1c 11.3%). A diagnosis of acute phlegmonous esophagogastritis with hypopharyngeal abscess was made based on typical computed tomography image features and clinical signs of infection. The patient received empirical antibiotic therapy initially; however, esophageal perforation with mediastinitis and empyema developed after admission. Emergency surgery with drainage and debridement was performed and antibiotics were administered. She was discharged in a stable condition on the 56th day of hospitalization. Six months later, a delayed esophageal reconstruction was performed. The patient has performed well for 9 months to date since the initial diagnosis.

Conclusions: Acute phlegmonous esophagogastritis complicated with hypopharyngeal abscess and esophageal perforation is extremely rare, and requires immediate medical attention. This report serves to remind physicians of this rare entity and the potential complications that may manifest with acute phlegmonous esophagogastritis.

MeSH Keywords: Abscess • Esophageal Perforation • Esophagitis • Gastritis

Full-text PDF: http://www.amjcaserep.com/abstract/index/idArt/902180

Background

Acute phlegmonous esophagogastritis is a rare digestive tract disease that has been reported only rarely in the literature [1–13]. Acute phlegmonous esophagogastritis is a potentially fatal disease that may be combined with esophageal perforation, mediastinitis, peritonitis, and empyema. Its typical presentation is intramural circumferential low attenuation surrounded by an enhanced rim of the esophageal and gastric wall. We present the classical radiological and endoscopic features and treatment strategy of a middle-aged female patient suffering from acute fulminant phlegmonous esophagogastritis complicated with hypopharyngeal abscess, esophageal perforation, mediastinitis, and empyema.

Case Report

A 60-year-old Taiwanese female presented with fever, fatigue, painful swallowing, and vague chest pain for 5 days. She was a homemaker with a past history of uncontrolled type 2 diabetes mellitus. On physical examination, general weakness, chest pain, odynophagia, and a fever up to 38.9°C were found, without signs of superficial localized infection. Respiratory rate and heart rate were within normal ranges, with no signs of sepsis initially. Positive laboratory results included leukocytosis (leukocyte count of 14.58×10^3/μL, neutrophils 76.8%) and serum glucose 348 mg/dL (HbA1c 11.3%). Computed tomography (CT) with contrast enhancement of the chest was performed, which revealed diffuse esophageal and focal gastric wall thickening with intramural circumferential low attenuation surrounded by an enhanced rim and hypopharyngeal abscess (Figures 1–3). A diagnosis of acute phlegmonous esophagogastritis with hypopharyngeal abscess was made based on typical computed tomography image features and clinical signs of infection, in addition to fatty stranding in the posterior mediastinum. Mediastinitis was also considered. Due to small perforations of the esophagus possibly not being depicted on the imaging study, an upper gastrointestinal panendoscopy was performed, which showed diffuse mural swelling of the esophagus without obvious esophageal perforation.

After admission, empirical antibiotic treatment with intravenous cefoxitin 2 g every 8 h was given. Intravenous esomeprazole 40 mg was administered once daily for stress ulcer prevention. Poor oral intake due to dysphagia and odynophagia was noted and total parenteral nutrition support was arranged. An insulin pump was prescribed for blood sugar control. Half-saline steam inhalation for sputum expectoration and enhanced chest care were performed 4 times daily.

On the 5th day after admission, the patient developed a cough with sticky sputum, and her oxygen saturation dropped to 93%

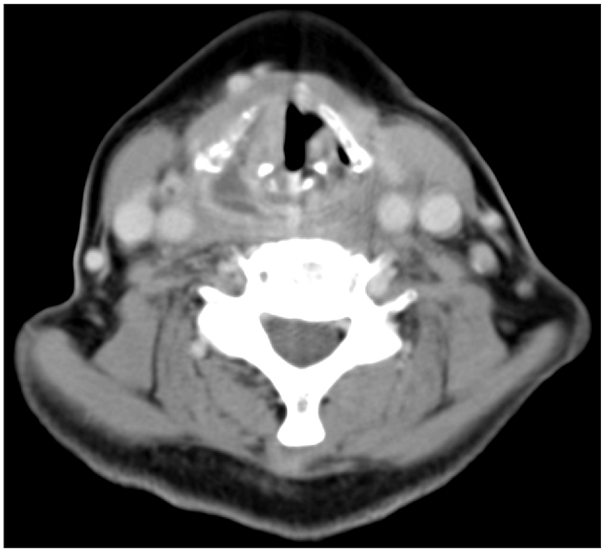

Figure 1. Contrast-enhanced computed tomography of the chest revealed hypopharyngeal abscess.

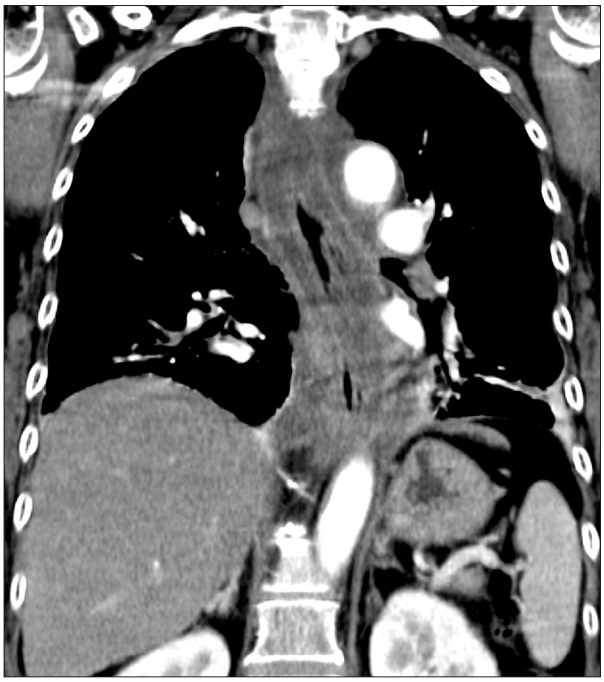

Figure 2. Contrast-enhanced computed tomography of the chest with coronal reconstruction demonstrated diffuse esophageal wall thickening.

on room air. A follow-up chest X-ray study revealed left massive pleural effusion. Laboratory investigations revealed leukocytosis, with white blood cell counts up to 27.6×10^3/μL (neutrophils 85.9%) and increased C-reactive protein (23.3 mg/dL). Emergency chest CT was performed, which demonstrated progression of the hypopharyngeal abscess and diffuse phlegmonous esophagogastritis with focal pneumatosis of the esophagus and pneumomediastinum, and esophageal perforation

Figure 3. Contrast-enhanced computed tomography of the chest with sagittal reconstruction showed diffuse esophageal and focal gastric wall thickening with circumferential intramural low attenuation surrounded by a peripheral enhanced rim. The intramural low attenuation represents severe inflammation and abscess localized to the submucosa and muscularis layer.

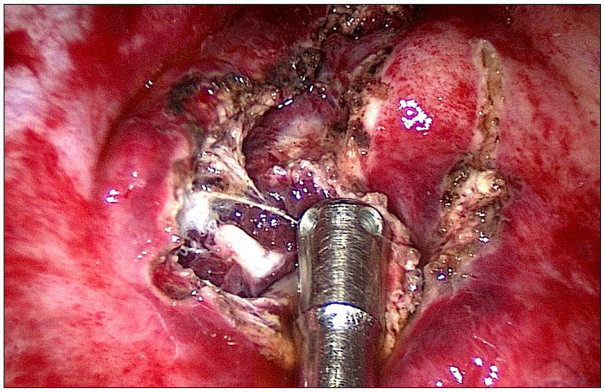

Figure 4. Video-assisted thoracic surgery demonstrated esophageal mural inflammation and necrosis with pus discharge.

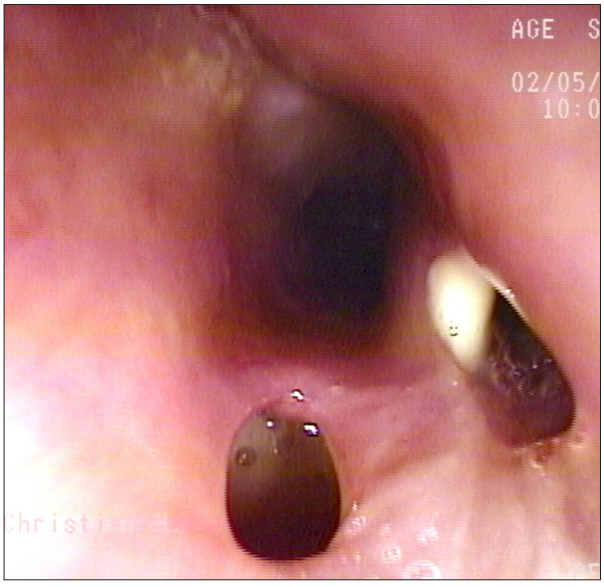

Figure 5. An upper gastrointestinal panendoscopy study revealed 2 holes in the esophageal wall, both about 3×3 mm in size, and at 19 cm from the incisors level, at 4 o'clock and 6 o'clock positions.

with a fistulous tract to the left pleural space with empyema. Emergency surgery consisting of decortication of the pleura by video-assisted thoracic surgery (Figure 4), mediastinotomy for debridement and drainage, and chest tube placement were performed, in addition to laparoscopic jejunostomy for nutrition supply. On the 8th day of hospitalization, enteral feeding via the jejunostomy tube was initiated. Sputum culture yielded *Pseudomonas aeruginosa*, and pleural effusion culture revealed *Klebsiella pneumoniae* and *Pseudomonas aeruginosa*. Antibiotic therapy was changed to imipenem 500 mg every 4 h and gentamycin 240 mg once daily.

On the 20th day of hospitalization, an upper gastrointestinal panendoscopy was performed, which revealed 2 holes in the esophageal wall, both approximately 3×3 mm in size at 19 cm from the incisors, at the 4 o'clock and 6 o'clock positions (Figure 5). Another hole approximately 1×1 mm in size was located 34 cm from the incisors, at the 4 o'clock position. Otherwise, esophagus swelling had improved in comparison with the previous examination. Surgery consisting of left cervical esophagostomy and right thoracoscopic pneumolysis, in addition to laparoscopic decompression gastrostomy, was performed. Extubation was completed on the 23rd day of hospitalization. In stable condition, the patient was transferred to an ordinary ward, and her postoperative course was uneventful. Subsequent follow-up chest CT showed a decrease in the extent of the previously-noted hypopharyngeal abscess and lesser esophageal and gastric wall thickening. Removal of chest tubes was performed on the 46th day of hospitalization. In stable condition and with adequate blood sugar control, the patient was discharged on the 56th day of hospitalization. Six months later, we performed esophageal reconstruction with colon interposition (terminal ileum, ascending colon, and transverse colon) via the retrosternal route, take-down of esophagostomy and

ileostomy, and closure of gastrostomy. The patient has done well during the first year (to date) since the initial diagnosis.

Discussion

Phlegmonous infection is a rare, life-threatening condition that may occur in the digestive tract. The stomach is the most commonly involved site, with more than 100 cases in the literature, reporting a mortality rate of 42% [14]. However, acute phlegmonous esophagitis is even rarer, with just a few cases reported (Table 1). Upon further review of the literature, we identified 2 case reports written in Japanese linking hypopharyngeal abscess to acute phlegmonous esophagogastritis [9,11]. However, these 2 cases were not complicated by esophageal perforation, mediastinitis, or empyema, and both described conservative treatment. To the best of our knowledge, our patient is the first documented case of acute phlegmonous esophagogastritis complicated with hypopharyngeal abscess, esophageal perforation, mediastinitis, and empyema in the English literature. Our case highlights the aggressive nature of acute phlegmonous esophagogastritis.

From the limited data presented in Table 1, we found that acute phlegmonous esophagogastritis primarily affects the middle-aged population, with a male predominance. The age of the previously-reported patients ranged from 31 to 73 years. The data included 10 males and 4 females. Due to the limited number of cases, the etiology of phlegmonous infection is unclear. However, some predisposing factors have been reported, including alcoholism, increased age, malnutrition with a low albumin level, low socioeconomic status, immunocompromised patients, tumor burden, and uncontrolled diabetes mellitus. More than one-third of the patients had uncontrolled diabetes mellitus as a predisposing factor, as was the case in our patient. All of the reported patients experienced the symptom of pain, either in the pharynx or in the chest. Dyspnea was noted as a symptom in 4 cases. *Klebsiella pneumonia* was the most common pathogen, which was present in more than one-third of the patients, although other pathogens have also been reported.

Acute phlegmonous esophagogastritis is a rare disease, and clinical diagnosis is often difficult due to the lack of pathognomonic signs or symptoms. Before modern diagnostic technology such as CT or endoscopy became available, a diagnosis of phlegmonous infection was rarely made before a surgery or an autopsy [12]. Endoscopic examination of an affected esophagus may demonstrate generalized luminal narrowing and poor distensibility and ulcers [15]. In our patient, a similar finding of diffuse mucosal thickening was observed in the first endoscopic examination.

CT image features are specific for acute phlegmonous esophagogastritis, with typical findings of an intramural circumferential low attenuation area of the esophagus and stomach surrounded by a peripheral enhanced rim after intravenous contrast medium administration. The intramural low attenuation is produced by an abscess in the submucosa and muscularis layer of the digestive tract [6,8]. If air bubbles are present within the thickened wall, infection caused by gas-forming pathogens is considered, and this image feature supports a diagnosis of acute phlegmonous esophagogastritis. In our patient, chest CT illustrated typical findings of acute phlegmonous infection with involvement of the hypopharynx, esophagus, and stomach. Due to extensive involvement and clinical signs of infection in the present case, diagnosis was definite. Image differential diagnoses such as intramural hematoma and tubular duplication of the esophagus exist, but there are no signs of infection in these conditions.

The reconstructive method of choice for most surgeons after esophagectomy is gastric interposition. Colon interposition is an appropriate alternative when the stomach is unavailable due to previous surgery, or malignant tumor. Concerns about using a gastric tube with gastrostomy include an additional possible site for postoperative leak or damage to the stomach, rendering it unusable. In our case, due to previous diffuse phlegmonous esophagogastritis, there is an additional potential concern of poor healing of cervical anastomosis if we choose the gastric interposition method. Therefore, we chose a colon interposition method to avoid complications.

Key factors in the control of acute phlegmonous infection as seen in our case are awareness of the disorder, early image diagnosis, antibiotics administration, surgical drainage, and adequate supportive care. Acute phlegmonous esophagogastritis may progress to esophageal perforation, empyema, peritonitis, and even death in reported gastritis cases [4,10,12,16]. A generally-accepted therapy for acute phlegmonous gastritis consists of systemic antibiotic treatment combined with partial or total gastrectomy [17]. However, due to the limited number of cases, a standard treatment for acute phlegmonous esophagogastritis has not yet been developed. In our patient, because there was no evidence of esophageal perforation nor peritonitis initially, therapy with broad-spectrum antibiotics was provided, and surgical intervention was not indicated. However, due to the development of esophageal perforation, mediastinitis, and empyema on the 5th day of hospitalization, decortication of the pleura, mediastinotomy for debridement and drainage, and chest tube placement were performed. The treatment strategy of this rare disease entity includes systemic antibiotic treatment to treat infection, prevention of progression of contamination, nutritional support, and preserving the continuity of the digestive tract, with timely surgical interventions if required [18–20].

Table 1. Summary of reported cases of phlegmonous esophagitis and the current case.

No./author (year)	Age (years) /sex	Risk factors	Symptoms	Involved organ	Pathogens	Tx	Major complications	Result
1/Mann et al. (1978) [1]	62/M	AL, ED	CP, DY, DS	E	EC, KP	C	Nil	A
2/Wakayama et al. (1994) [2]	31/M	Nil	CP	E, S	BS, EC, KP	C	Nil	A
3/Hsu et al. (1996) [3]	42/M	Nil	CP, F, DY	E, S	GPB	S	Hematemesis	A
4/Furuchi et al. (1998) [4]	49/M	Tonsillitis	CP	E, S	Not detected	S	Peritonitis	A
5/Kawakubo et al. (2002) [5]	51/M	Nil	CP	E, S	Not detected	C	Nil	A
6/Jung et al. (2003) [6]	52/M	DM	CP	E, S	GPB	C	Esophageal ulcers	A
7/Tsukadaira et al. (2005) [7]	41/M	Dental caries	CP, F	E, S, D	*	C	Nil	A
8/Yun et al. (2005) [8]	63/F	DM	CP	E	GPB	C	Nil	A
9/Imai et al. (2005) [9]	73/F	Nil	CP	H, E, S	AS	C	Nil	A
10/Nishiya et al. (2007) [10]	43/M	AL	F, DS	E	KP	C	Esophageal stenosis and perforations	A
11/Shiozawa et al. (2009) [11]	62/M	DM	CP	H, E, S	AS	C	Nil	A
12/Kim et al. (2010) [12]	48/M	AL, DM	CP, DY	E, S	KP	S	Empyema	A
13/Karimata et al. (2014) [13]	47/F	CRT	CP, DY	E	SM	C	Nil	A
14/The current case	60/F	DM	CP, F, DS, OD	H, E, S	KP, PA	S	Esophageal perforations, mediastinitis, empyema	A

Tx – treatment; AL – alcoholism; ED – epiphrenic diverticulum; DM – diabetes mellitus; CRT – chemoradiotherapy; CP – chest pain; DY – dyspnea; DS – dysphagia; F – fever; OD – odynophagia; E – esophagus; S – stomach; D – duodenum; H – hypopharynx; EC – *Enterobacter cloacae*; KP – *Klebsiella pneumoniae*; GPB – Gram-positive bacilli; * – *Peptostreptococcus micros*, *Fusobacterium* sp., a-*Streptococcus*, *Gemella morbillorum*; AS – a-*Streptococcus*; SM – *Streptococcus milleri*; PA – *Pseudomonas aeruginosa*; C – conservative; S – surgery; A – alive.

Conclusions

Acute phlegmonous esophagogastritis complicated with hypopharyngeal abscess and esophageal perforation is extremely rare and requires immediate medical attention. Clinical symptoms are nonspecific, and early radiologic diagnosis is key to achieve a better prognosis. This report serves to remind physicians of this rare entity and the potential complications that may manifest with acute phlegmonous esophagogastritis.

Acknowledgements

The authors thank the Department of Chest Surgery for patient care.

Statement

The authors declare that they have nothing to disclose.

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