

# Acute Phlegmonous Esophagogastritis Causing Respiratory Distress

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## ABSTRACT

Phlegmonous esophagogastritis is an extremely rare condition characterized by diffuse inflammation of the upper gastrointestinal tract, sparing the mucosa. Patients can present with an acute onset of symptoms, and computed tomography scans can show diffusely edematous wall with intramural low attenuation surrounded by ring enhancement. Here we report such a case of a man who presented with central chest pain and breathing difficulty. The patient developed respiratory distress due to compression of trachea by the edematous esophagus.

## INTRODUCTION

Acute phlegmonous esophagogastritis is a disease of the digestive tract that has been reported rarely in literature. It is a potentially fatal disease and requires early diagnosis and aggressive management. This is an infectious condition and high mortality is due to difficulty in source control. Radiological presentation is typical with intramural circumferential low attenuation area surrounded by ring enhancement of esophageal and gastric wall. From the available literature, the mortality rate is very high, even with surgical interventions.

## CASE REPORT

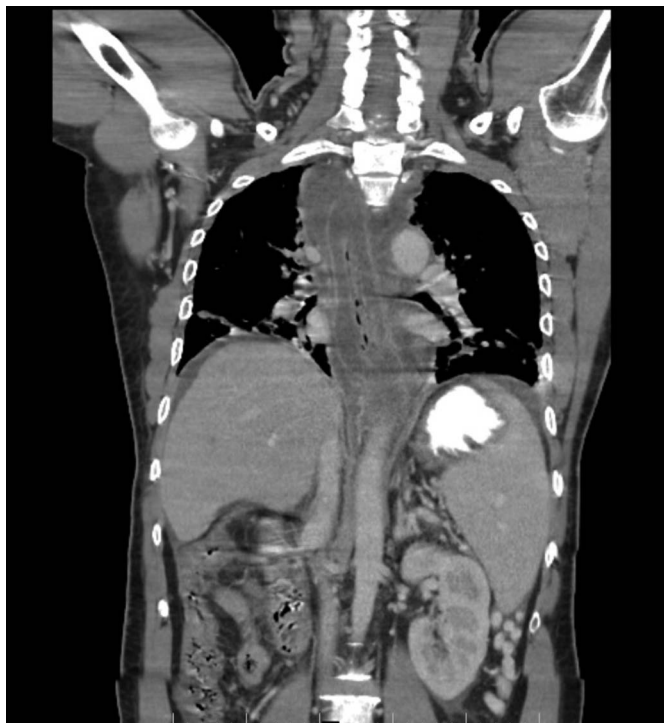
A 56-year-old man with uncontrolled diabetes (HbA1C: 7) and a history of occasional alcohol consumption presented with central chest pain and breathlessness for 5 days. There was associated fever, dysphagia, and abdominal discomfort with belching, for which he took antacids, but there was no relief. There was no history of vomiting or loose stools. He was evaluated in a local hospital where he was referred for endoscopy. The next day, before the procedure, the patient developed severe respiratory distress with noisy breathing, dyspnea, and desaturation after which he was intubated and shifted to our hospital for further management.

On admission, the patient was intubated and sedated. His heart rate was 121 beats per minute, respiratory rate was 19 breaths per minute, and blood pressure was 100/80 mm Hg, with oxygen saturation of 100% in 30% FiO<sub>2</sub> with a temperature of 100°F. Respiratory system and abdominal examination was unremarkable. On admission, his total white blood cell count was 12,000/UL with 85% neutrophils, C-reactive protein was 171 mg/L, and creatinine was 1.6 mg/dL. Arterial blood gas showed metabolic acidosis with lactate of 5.6 mmol/L. His computed tomography (CT) scan of thorax, which was done before intubation, revealed diffusely edematous esophagus and lesser curvature of stomach (Figures 1 and 2) with minimal collection around upper esophagus and fat stranding, suggestive of mediastinitis. He was started on piperacillin—tazobactam and other supportive medication. Other organs including lung parenchyma were normal in CT. There was significant compression of trachea by the edematous esophagus. Upper gastrointestinal scopy showed normal mucosa (Figure 3), and biopsy taken from gastric body showed normal epithelium, lamina propria, and cellularity. CT findings were typical of phlegmonous esophagogastritis. Gradually, the patient's condition worsened with sepsis, and he was started on multiple inotropic supports. Two blood cultures showed growth of *Klebsiella pneumoniae*. His total blood count increased to 25,000 and renal functions worsened with a creatinine of 1.9. In view of his deteriorating condition, exploratory thoracotomy was performed. Pus collection was found around upper esophagus just above the azygos vein. Mediastinal pleura along the whole length of the esophagus lay open and pus was let out. Postoperatively, the patient's condition remained the



**Figure 1.** Contrast computed tomography of thorax showing edematous esophagus with low attenuation area, surrounded by ring enhancement (arrow) and adjacent pus collection. Severely compressed trachea due to edematous esophagus.

same and there was pus drainage through chest tubes. Antibiotics were upgraded to meropenem because blood and pus culture showed *Klebsiella*. However, despite all supportive measures, his condition deteriorated and the patient died on the fifth day of hospital admission.



**Figure 2.** Contrast computed tomography of thorax-coronal section showing diffuse thickening of esophagus (arrow) with mediastinal fat stranding.

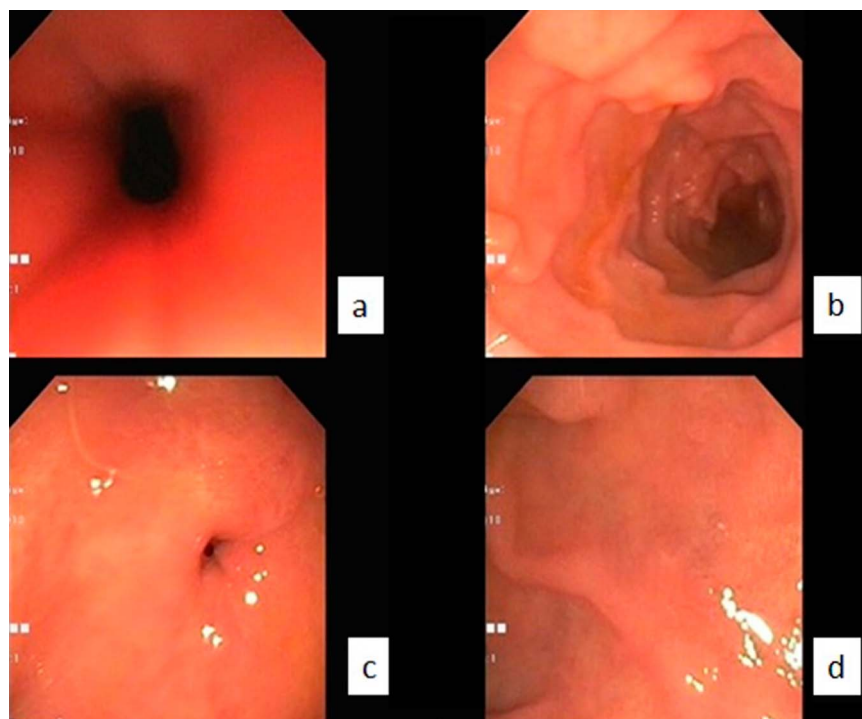
## DISCUSSION

Phlegmonous esophagogastritis is a rare entity with high mortality and only few case reports are available. Phlegmon is an inflammatory process spreading diffusely associated with exudate or pus formation.<sup>1</sup> Phlegmonous infection can involve any part of gastrointestinal tract, the most common site being the stomach.<sup>2</sup> Other sites in reported cases include esophagus, small bowel, colon, appendix, etc.<sup>1-4</sup> Phlegmonous infection of both stomach and esophagus is extremely rare. Here, we report a case of phlegmonous esophagogastritis that caused severe tracheal compression and respiratory distress. To our knowledge, this is the only case having such a presentation.

Phlegmonous infection usually involves submucosa and not mucosa.<sup>1,3,4</sup> Inflammation can involve submucosa, muscularis mucosa, and serosa. Because of severe inflammation, these layers become edematous and CT shows a radiolucent area, which is well demarcated by serosa and mucosa. Phlegmonous gastritis is more commonly seen than phlegmonous esophagitis. Localized form of phlegmonous gastritis usually affects gastric antrum.<sup>5</sup> Diffuse form involves the entire stomach and rarely extends beyond cardia or pylorus of the stomach.<sup>6</sup> Endoscopy may show luminal narrowing, mucosal fold thickening, or ulcer-like lesions.<sup>1</sup> Endoscopic ultrasonography can show diffuse thickening of submucosal layer with hypoechoic areas.<sup>1,6</sup> In our case, endoscopy revealed absolutely normal mucosa and even biopsy at the level of lesser curvature was normal.

Exact etiology of phlegmonous esophagogastritis is unclear. The most common factors in reported cases are immune suppression, alcoholism, peptic ulcer disease, infection, trauma, foreign body aspiration, and peptic ulcer disease.<sup>4,7</sup> Our patient was a diabetic and had significant alcohol ingestion. These factors may have predisposed the entry and growth of pathogenic organisms by altering the body's defense mechanisms. In more than half of the reported cases, patients were healthy previously and there were no predisposing factors.<sup>8</sup> In our case and in most of the reported cases, symptoms had an acute onset followed by rapid deterioration.<sup>2</sup> In many cases, postmortem biopsy showed thickened submucosa, infiltrated with inflammatory cells, exudate, hemorrhage, necrosis, and thrombosis of submucosal blood vessels.<sup>1,2</sup> The most common organisms were *Streptococcus*, *Staphylococcus*, *Escherichia coli*, *Haemophilus influenzae*, and *Klebsiella*. In our case, the organism was *Klebsiella* based on culture growth in the blood and pus samples that were aspirated during surgery.

From previous case reports, acute phlegmonous esophagogastritis primarily affects middle-age population, with male preponderance.<sup>4</sup> Common symptoms during presentation were fever, chest pain, nausea, vomiting, hematemesis, hiccups, and dysphagia. In many cases, dyspnea was due to associated pleural effusion. However, in our case, pleural involvement was not there at initial presentation, and dyspnea was due to tracheal



**Figure 3.** Endoscopic image of esophagus (A), pylorus (B), antrum (C), and body (D) of stomach.

compression. Cases with tracheal compression causing stridor were never reported. Most of these cases were diagnosed at surgery or during autopsy.

The characteristic CT findings of phlegmonous esophagogastritis include diffuse wall thickening of esophagus and stomach with circumferential intramural low attenuation surrounded by a peripheral ring enhancement.<sup>1</sup> The low attenuation area represents severely inflamed area and abscess localized by the submucosa and muscularis layer.<sup>9,10</sup> Clinical and radiological features of different reported cases are compared in Table 1.

Radiological differential diagnosis include dissecting intramural hematoma, tubular duplication of esophagus, and emphysematous esophagitis.<sup>9,11,12</sup> In dissecting intramural hematoma, chest pain will be the predominant symptom and association of infection and sepsis are unlikely. In tubular duplication, patients are usually asymptomatic with no signs.

Kim et al<sup>6</sup> showed mortality of 20% in patients who underwent surgical resection and 50% in those who were managed conservatively with an overall mortality of 42%. Among reported cases, those who were managed surgically had a better

Author	Kim et al <sup>1</sup>	Huang et al <sup>4</sup>	Jung et al <sup>9</sup>	Karimata et al <sup>17</sup>
Condition	Phlegmonous esophagogastritis	Phlegmonous esophagogastritis	Phlegmonous esophagogastritis	Phlegmonous esophagitis
Age (yr)	48	60	52	47
Sex	Male	Female	Male	Female
Comorbidity	Alcoholism, diabetes mellitus	Diabetes mellitus	Diabetes mellitus	Carcinoma cervix on chemotherapy
Symptoms	Left chest pain, dyspnea, abdominal pain: 5 d	Fever, dysphagia, chest pain: 5 d	Dysphagia, fever: 2 d	Vomiting, epigastric pain: 1 d
Endoscopy	Esophageal ulcer, gastric mucosal hemorrhage	Diffuse mural swelling of esophagus without perforation	Normal, except pus in pyriform sinus	Normal mucosa
Radiology	Diffuse circumferential wall thickening of esophagus, gastric cardia. Bilateral pleural effusion	Same with hypopharyngeal abscess, mediastinitis	Same with abscess in area of pyriform sinus	Diffuse thickening of esophagus, bilateral pleural effusion
Organism	<i>Klebsiella</i>	<i>Klebsiella</i> + <i>Pseudomonas</i>	Gram-positive bacilli	<i>Streptococcus milleri</i>
Management	Surgical debridement	Surgical debridement	Antibiotics alone	Antibiotics alone

prognosis. Surgical drainage, appropriate antibiotics, and supportive measures are the mainstay of the treatment. Possible complications of phlegmonous esophagogastritis are sepsis, esophageal ulceration, necrosis, mediastinitis, and peritonitis. Long-term complications include esophageal stricture, gastric mucosal atrophy, etc.<sup>2,13</sup>

Because of the limited number of cases, a standard treatment for phlegmonous esophagogastritis has not yet been developed. Some surgeons prefer surgical treatment with partial or total gastrectomy for phlegmonous gastritis. But there are many case reports of successful treatment with antibiotics alone.<sup>14</sup> The treatment strategy should include systemic antibiotics, prevention of the progression of contamination, maintaining the continuity of digestive tract, nutritional support, and timely surgical intervention when required.<sup>4,15,16</sup> In our case, even with good antibiotic cover and surgical drainage, there was no good response, and finally, the patient died from sepsis.

In conclusion, phlegmonous esophagogastritis is a rare condition and it can present with respiratory distress due to tracheal compression. Patients need early diagnosis and aggressive management in view of high mortality.

## DISCLOSURES

Author contributions: R. KO collected the data, wrote the manuscript, reviewed the literature, and is the article guarantor. J. Baby wrote the manuscript and revised the literature. P. Valsalan K wrote the manuscript and reviewed the literature.

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Informed consent obtained from patient's next of kin.

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## REFERENCES

- Kim HS, Hwang JH, Hong SS, et al. Acute diffuse phlegmonous esophagogastritis: A case report. *J Korean Med Sci*. 2010;25(10):1532-5.
- Hsu CY, Liu JS, Chen DF, Shih CC. Acute diffuse phlegmonous esophagogastritis: Report of a survived case. *Hepatogastroenterology*. 1996;43(11):1347-52.
- Wakayama T, Watanabe H, Ishizaki Y, et al. A case of phlegmonous esophagitis associated with diffuse phlegmonous gastritis. *Am J Gastroenterol*. 1994;89(5):804-6.
- Huang YC, Cheng CY, Liao CY, Hsueh C, Tyan YS, Ho SY. A rare case of acute phlegmonous esophagogastritis complicated with hypopharyngeal abscess and esophageal perforation. *Am J Case Rep*. 2017;18:125-30.
- Jung C, Choi YW, Jeon SC, Chung WS. Acute diffuse phlegmonous esophagogastritis: Radiologic diagnosis. *Am J Roentgenol*. 2003;180(3):862-3.
- Kim GY, Ward J, Hennessey B, et al. Phlegmonous gastritis: Case report and review. *Gastrointest Endosc*. 2005;61(1):168-74.
- Stein LB, Greenberg RE, Ilardi CF, Kurtz L, Bank S. Acute necrotizing gastritis in a patient with peptic ulcer disease. *Am J Gastroenterol*. 1989;84(12):1552-4.
- Kim GY, Ward J, Hennessey B, et al. Phlegmonous gastritis: Case report and review. *Gastrointest Endosc*. 2005;61(1):168-74.
- Jung C, Choi YW, Jeon SC, Chung WS. Acute diffuse phlegmonous esophagogastritis: Radiologic diagnosis. *AJR Am J Roentgenol*. 2003;180(3):862-3.
- Yun CH, Cheng SM, Sheu CI, Huang JK. Acute phlegmonous esophagitis: An unusual case (2005: 8b). *Eur Radiol*. 2005;15(11):2380-1.
- Jung JH, Choi HJ, Yoo J, Kang SJ, Lee KY. Emphysematous gastritis associated with invasive gastric mucormycosis: A case report. *J Korean Med Sci*. 2007;22(5):923-7.
- van Mook WN, van der Geest S, Goessens ML, Schoon EJ, Ramsay G. Gas within the wall of the stomach due to emphysematous gastritis: Case report and review. *Eur J Gastroenterol Hepatol*. 2002;14(10):1155-60.
- I H, Park CS, Kim YD. Treatment of phlegmonous esophagitis combined with mediastinitis. *Korean J Thorac Cardiovasc Surg*. 2007;40(10):711-4.
- Iwakiri Y, Kabemura T, Yasuda D, et al. A case of acute phlegmonous gastritis successfully treated with antibiotics. *J Clin Gastroenterol*. 1999;28(2):175-7.
- Iwakiri Y, Kabemura T, Yasuda D, et al. A case of acute phlegmonous gastritis successfully treated with antibiotics. *J Clin Gastroenterol*. 1999;28(2):175-7.
- Isik A, Firat D, Peker K, Sayar I, Idiz O, Soytürk M. A case report of esophageal perforation: Complication of nasogastric tube placement. *Am J Case Rep*. 2014;15:168-71.
- Karimata H, Nishimaki T, Oshita A, et al. Acute phlegmonous esophagitis as a rare but threatening complication of chemoradiotherapy: Report of a case. *Surg Today*. 2014;44(6):1147-51.

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