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Case Report

Two-stage surgery for delayed esophageal perforation and concomitant chylothorax secondary to upper gastrointestinal endoscopy

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ABSTRACT

A 46 years old male smoker was admitted to our hospital with a three-month history of chest discomfort and burning sensations due to regurgitation of food. The gastroenterologist tried multiple attempts to pass the endoscope through the lower end of the esophagus but failed. Post endoscopy Chest -X-ray showed right hemithorax fluid collection. A 28Fr chest drain was inserted, and fluid analysis revealed chyle. A contrast computed tomographic scan of the chest (CT) revealed esophageal perforation. The patient was managed conservatively by the primary physician on TPN, Antibiotics, and keeping him nil by mouth. After two weeks of failed conservative management, they referred the patient to the thoracic surgeon. We planned two-stage surgery because the patient was critically sick, septic, and hemodynamically unstable on inotropic support.

1. Background

Esophageal perforation signs and symptoms are misleading, leading to difficulty in timely diagnosis and appropriate management of this devastating clinical entity. The iatrogenic injuries have surpassed the other causes due to recent advances in endoscopic procedures [1]. Early diagnosis and management have a crucial role in the outcome. Barrett et al., in 1946, was the first to report a case of spontaneous esophageal perforation [2]. Hermann Borhaaves 1723 described the complete esophageal rupture after conducting the post mortem of the High Admiral of the Dutch Navy, Baron van Wassenaar. He was found dead in his room due to intense and prolonged vomiting after excessive ingestion of food and alcohol at a late-night party [3]. In 1947 Barrett Claggett was the first to report a successful surgical repair following esophageal perforation [4].In 1952 Satinsky and Kron performed a successful esophagectomy following perforation [5].In recent years mortality due to esophageal perforation has markedly declined as compared to the last century due to advances in diagnostic modalities and surgical approaches and the availability of broad-spectrum antibiotics. This case is reported in line with scare criteria [6].

2. Case report

A 45 years old male smoker known case of hypertension and hepatitis C presented with a history of mild hematemesis. Past medical history of drug abuse (cannabis and amphetamine). The clinical examination was unremarkable. Upper gastrointestinal endoscopy (UGIE)was planned to rule out the cause of hematemesis. Gastroenterologists failed to pass the scope through the lower end of the esophagus despite multiple attempts with different sizes of an endoscope. A chest X-ray showed a large right pleural effusion the next day and was drained by inserting a size Fr28 chest drain. Initial fluid was brownish in color; later on, it became milky, and daily output was from 1.5 to 2.0 L/day Fig. 1(A). On day three patient became hypotensive and had shortness of breath. The CT scan of Thorax with contrast showed leakage of contrast into the chest cavity. Fig. 1(B,C&D)

The patient was managed non-operatively by the primary physician, keeping him nil by mouth, and he started Total parental nutrition (TPN) systemic broad-spectrum antibiotics. There was no improvement, and the patient's condition deteriorated; he lost a lot of weight, became septic, hypotensive, and had breathing difficulty. He was moved to the Intensive care unit (ICU). He was losing almost 2 L of chyle/day. This case was referred to a thoracic surgeon two weeks after the perforation. The pleural fluid culture was positive for Enterobacter Cloacae S: Bactrim. Blood culture was positive for candida and staphylococcus aureus. Systemic antibiotics and antifungals were started (Meropenum 1Gm six-hourly, Tazobactam 4.5 Gm x six-hourly, Vancomycin 500mgx12 hourly, Fluconazole 200mg daily, caspofungin 50mg daily). The

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experienced thoracic surgeon decided to proceed with life-saving two-stage surgery in this delayed esophageal perforation and persistent chyle leak because the patient was critically sick, septic, and immune-compromised due to a prolonged chyle leak. We did stage one surgery through a right thoracotomy, ligated the thoracic duct, and debridement and lung decortication were performed. After washing the Chest cavity, we noticed anterior and posterolateral mid esophageal perforations extending distally up to the gastroesophageal junction. The perforated part of the esophagus was completely covered with a thick pleural patch to avoid further soiling the chest cavity. A nasogastric feeding tube was placed up to 3rd part of the duodenum. The patient was transferred to ICU. Inotropes were weaned off, and TPN and broad-spectrum antibiotics and antifungals were continued. The next day NG feed was started and gradually built up to improve intestinal mucosal functional integrity. The patient's general condition improved, and he was mobilized. On day ten, after the first stage surgery, we performed second stage surgery firstly through laparotomy stomach was mobilized, and the gastric tube was created. Then through a right thoracotomy, after a very difficult dissection lower esophagus was mobilized and perforated part of the esophagus was removed, and anastomosis was done above the azygous vein Fig. 2(A and B,C&D). Feeding jejunostomy was fashioned, and the patient was transferred to ICU and later on to the ward. On day six, after the second stage, chest x-ray and Gastrografin swallow showed no leak Fig. 3(A,B&C). Oral fluid intake was started and gradually built up to solids. A chest drain was removed, and the patient was discharged home for a follow-up in outpatient.

3. Discussion

Esophageal perforation is a most dreadful complication of endoscopic procedures with a high rate of mortality and morbidity. Although esophageal perforation has been uncommon over recent years, its prevalence is increasing worldwide because of the widespread use of endoscopy for diagnostic and therapeutic purposes. Iatrogenic injury to the esophagus has surpassed the other causes of esophageal perforation, instrumentation accounting for 59%, and Spontaneous perforations accounting for 15%. Other injuries included foreign body ingestion (12%), trauma (9%), operative injury (2%), tumor (1%), and other causes (2%) [7,8]. Incidence of Esophageal perforation during upper gastrointestinal endoscopy (UGIE) is approximately 0.03% and 0.11% during flexible and rigid endoscopy, respectively. Iatrogenic esophageal perforation is usually encountered at the normal anatomic narrowings of the esophagus. The cervical esophagus perforation results due to a forceful attempt of passing the endoscope through the cricopharynx [9].

Lower esophagus perforation is most frequently encountered when esophageal dilation is performed for esophageal strictures or achalasia. The incidence of lower esophageal rupture after pneumatic dilation for achalasia varies from 2% to 6% [10]. Endoscopic sclerotherapy for esophageal varices leads to esophageal perforation in 1%–3% of patients [11]. Salo et al. reported incidence of esophageal perforation is 1–3% after endoscopic sclerotherapy, which is due to transmural necrosis of the esophageal wall [12]. Daniel et al. reported that the incidence of esophagus perforation during echocardiography (ECHO) is 0.18% [13].

A rare cause of lower esophagus rupture is the use of Sengstaken-Blakmore to control bleeding esophageal varices, nasogastric tube

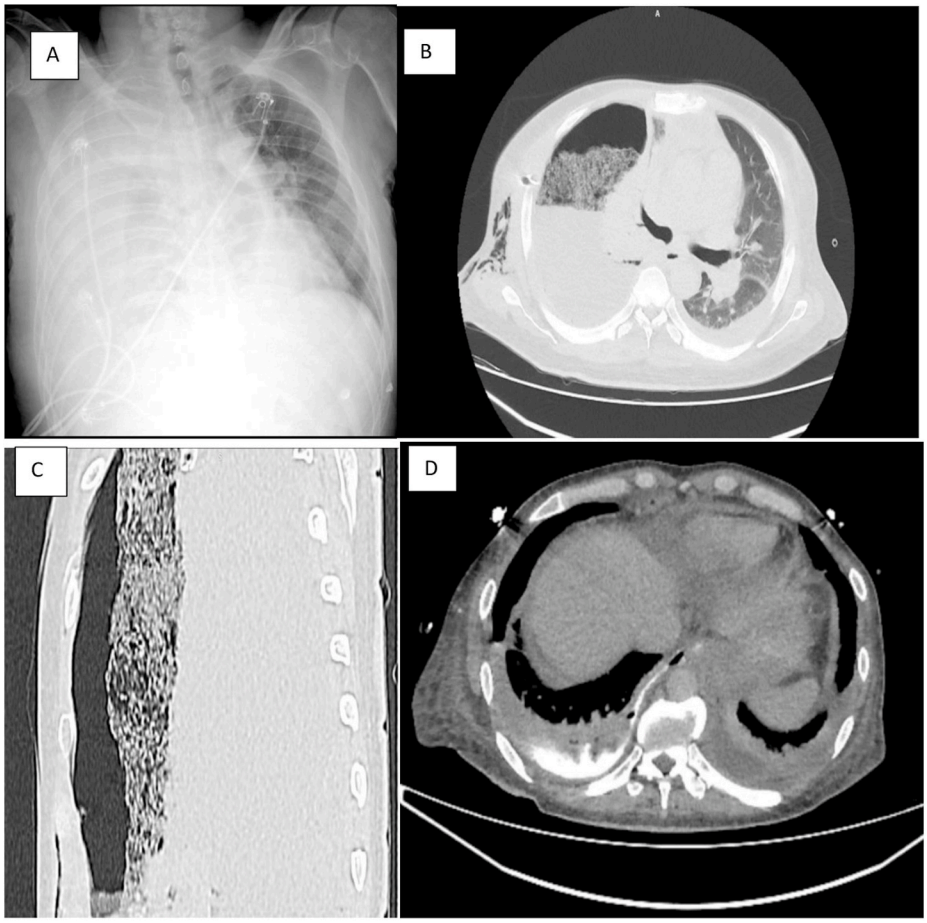

Fig. 1. (A) Post endoscopy Chest X-Ray showing a opacification of right hemithorax (Large pleural effusion).
(B)CT scan of chest axial view showing large pleural effusion and chyl froth. (C)CT Scan chest Sagittal view showing lung collapse and chyle layer. (D)Contrast CT scan of chest showing leak of contrast in to the pleural space confirming esophageal perforation.

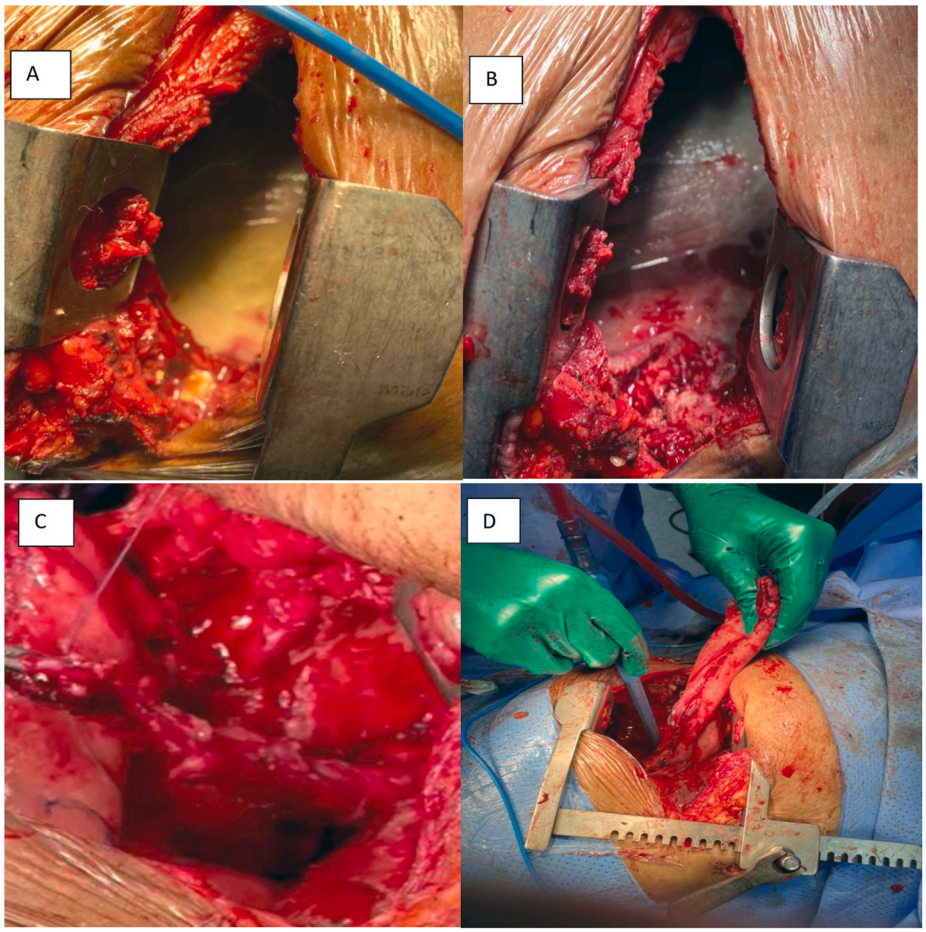

Fig. 2. (A)Right Thoracotomy showing chyle and trapped lung. (B)Thoracic duct ligated and clipped. (C) esophagus with multiple perforations. (d) Stomach conduit.

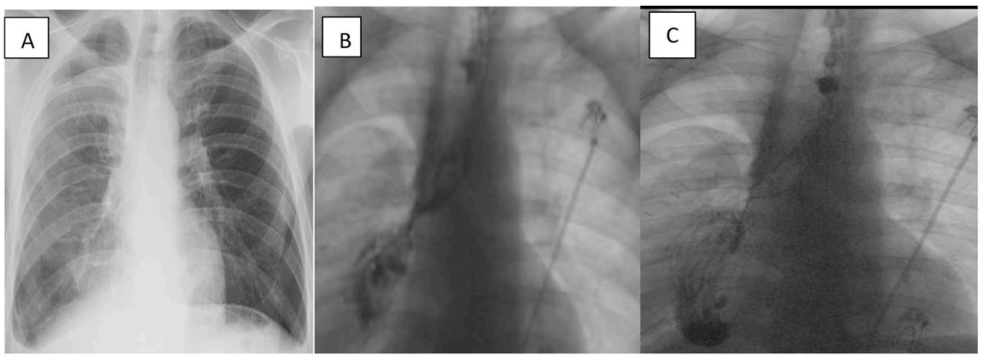

Fig. 3. (A) Chest X-Ray on discharge. (B) Gastrografin study showing free flow of contrast no leak patent anastomosis (C) Contrast in gastric conduit.

insertion, esophageal intubation, endotracheal tubes, endoscopic retrograde cholangiopancreatography, and endoscopic ultrasound-guided interventions [14–17].

The clinical manifestations of early esophageal injury are usually vague and nonspecific. Therefore, every effort should be made to avoid delays in establishing an accurate diagnosis to avoid the contamination of mediastinum. The clinical presentation varies with the cause, site, size, and time period elapsed after injury of perforation. The delay in the diagnosis leads to the soiling of the mediastinum and high mortality [18,19].

Nesbitt and Sawyers et al. reviewed the Clinical presentation of esophageal injuries during a 50-year period and found pain to be the most common symptom (71%), followed by fever (51%) dyspnea (24%), and crepitus (22%) [20].

Patients with thoracic esophageal perforation usually present with retrosternal or chest pain lateralizing to the side of perforation.

The visceral mediastinum is initially contaminated with leaked saliva and its enzymes and gastric contents. This initiates the intense inflammatory response and cytokine activation leading to tissue necrosis and severe mediastinitis, perforation of mediastinal pleura fluid sequestration, sepsis, and hemodynamic instability. Right and left pleural spaces are contaminated by the upper and distal esophageal perforations, respectively [20,21,22]. The intraabdominal esophageal perforation commonly presents as a dull epigastric pain radiating to the back, while the anterior perforation may present as severe epigastric pain, fever, sepsis, peritonitis, fever, and other systemic signs.

The diagnosis of this lethal condition can be very difficult and often misleading, leading to delays in management. Due to the relative rarity and nonspecific presentations, delay in the diagnosis and management has been reported in more than 50%esophageal perforations [23]. The chest x-ray in the early phase may not show any abnormality. If on Chest X-ray there is a finding of subcutaneous emphysema, pleural pneumothorax, pleural effusion pneumomediastinum, hydropneumothorax, subdiaphragmatic air, there is a strong possibility of esophageal rupture [24].

Panzini et al. conducted a retrospective study about chest x-ray findings in instrumental esophageal perforations found 80% of the patients had abnormalities, pneumomediastinum 60%, and density in left cardio phrenic angle 33% with a loss of descending aorta contour [25]. The gold standard diagnostic tool is still the contrast esophagography using a water-soluble contrast agent (Gastrografin). The contrast extravasation is noted in 50% of cervical and 75–80% of esophageal perforations. If this fails to detect esophageal perforation, then alternatively, a dilute barium swallow can be very useful to detect any leak. Almost 90% of thoracic esophageal perforation is detected by this imaging modality [26–28].

A contrast CT scan is very useful in case there is any doubt about the diagnosis. In addition to this, a drainage procedure can be executed at the same time. CT scan abnormal findings suggestive of esophageal perforations are pneumomediastinum, esophageal thickening, communication between air-filled esophagus and contiguous mediastinal and Para mediastinal air-fluid collection, or abscess adjacent to the esophagus. Left side pleural effusion or hydropneumothorax is seen in distal esophagus perforation, while these findings are commonly seen on the right side due to middle esophagus perforations [29,30]. Pleural fluid analysis may show undigested food, pH less than 6.0, and elevated levels of salivary amylase, which confirms the diagnosis [31].

Esophageal perforation and concomitant chylothorax due to UGIE have not been reported in the medical literature before. Chylothorax has been reported after esophagectomy, endoscopic sclerotherapy, and endoscopic ultrasound-guided esophageal biopsy [32,33].

The critical prognostic factor for the successful outcome of esophageal perforation is the etiology, site, severity of perforation, and time period between diagnosis, initiation of management, and perforation. Other factors such as general health of the patient, comorbidities, sepsis, and necrosis of mediastinal tissues are also important. The main aim of treatment is drainage of pleural space, avoiding further contamination and infection due to perforation, intravenous antimicrobial therapy, and restoration of the integrity of the gastrointestinal tract with nutritional support [32]. These patients are at risk of developing sepsis, respiratory failure, shock, and mediastinal tissue necrosis. Therefore treatment should be tailored according to the need of the patient as per his clinical condition. Treatment options include non-surgical or Surgical. Nonoperative management has a limited role in selected patients with well-contained leaks and minimal pleural mediastinal contamination [33]. Cameron et al. proposed a nonoperative treatment should be considered if perforation is contained and drained back into the esophagus, symptoms are mild, and there is minimal evidence of sepsis [34]. Mongol and Klassen 1965 described nonoperative management in 18 patients with esophageal perforation that were diagnosed within 24 hours. Only one death was reported [35]. Brinster et al. reported a retrospective analysis of 559 patients with esophageal perforation managed non-operatively and 322 managed with primary surgical repair, and the mortality was 18% and 12%, respectively [36].

Nonoperative treatment requires a diligent assessment and is safe in selective patients. Altorjay et al. and other authors established a guideline for the non-surgical treatment [37,38].

* Early diagnosis or leak contained if the diagnosis is delayed.
* Leak contained within neck or mediastinum or between the mediastinum and visceral lung pleura.
* Drainage into the esophageal lumen as evidenced by contrast imaging.
* Injury not in neoplastic tissue, not in the abdomen, and not proximal to the obstruction.
* Symptoms and signs of septicemia absent and
* Contrast imaging and experienced thoracic surgeon available

Nonoperative management includes keeping the patient nil by mouth for 2–3 days, nasogastric tube, broad-spectrum antimicrobial therapy, total parenteral nutrition, and drainage of pleural or mediastinal collection by chest drain or CT guided catheters [39,40]. Although few cases have been reported successfully treated with endoscopically placed coated stents and clipping, their clear role yet needs to be established. The major drawback of nonoperative treatment is prolonged drainage, esophageal diversion, mediastinitis, necrotic esophageal and Para esophageal tissues, and persistent leak. Reconstructive surgery in such cases is very difficult [41–43].

Operative management includes primary closure with or without autogenous tissue reinforcement, exclusion and diversion, esophageal T tube, and esophagectomy [44].

Although Berrett, Olsen, and Claggett reported primary repair of esophageal perforation sixty years ago, it is still the gold standard of treatment for the esophageal perforation, provided esophageal tissues are repairable mucosal edges are healthy after necrosectomy. There should be no concomitant distal obstruction, and the defect is less than one-third of the esophagus circumferential diameter [45].In reinforced primary repair, a variety of autogenous tissues like diaphragm pedicle flap, rhomboid and latissimus dorsi, intercostal muscles, pleura, and pericardium has been used to augment the primary repair. Results of the primary repair and reinforced repair are almost similar. Wright and associates reported overall mortality of 14% after reinforced primary repair for esophageal perforation [46–49]. Whyte et al. also reported the same results after primary repair in a group of 22 patients [50]. Maghissi and Pender said mortality of 100% if there is obstruction distal to primary repair, while the mortality is 29% if there is no distal obstruction [51]. Mortality due to esophagus perforation has markedly decreased over the years from 60% to 10–14% due to early diagnosis by modern diagnostic modalities and advanced surgical skills. Two-stage surgery is a useful surgical technique for delayed esophageal perforation with concomitant chylothorax in a critically sick patient.

Esophageal perforation and concomitant chylothorax due to UGIE have not been reported in the medical literature before. Chylothorax has been reported after esophagectomy, endoscopic sclerotherapy, and endoscopic ultrasound-guided esophageal biopsy[52,53].

The incidence of iatrogenic chylothorax is 0.5 %–3% and has dominated the other causes with recent advances in medical procedures. Untreated chyle leak can lead to cardiorespiratory, hemodynamics instability, malnutrition, and immunosuppression. Early intervention is warranted if the chyle leak is $>$ 1.5-L day or is more than 1 L/day for five days. The mortality from untreated chyle leak is 20%–60%. Chyle leak after esophagectomy occurred in 10% of cases, and reported mortality is 50% [54,55].

In our case, multiple mid-esophageal perforations extended to the distal esophagus. Probably thoracic duct was also damaged during the endoscopy. Late diagnosis and conservative management further delayed the surgery. As the patient was critically ill, immune-compromised, septic, malnourished, decreased muscle mass, and hypotensive on inotropic support noradrenaline 15 μg. Two-stage surgery was planned and successfully managed for the patient with excellent results.

4. In conclusion

We are reporting the first case of delayed Esophageal Perforation and concomitant Chylothorax secondary to upper Gastrointestinal endoscopy. A patient with late-diagnosed esophageal perforation and

concomitant chylothorax is critically ill and cannot withstand a major surgical procedure of gastric pull-up and esophagectomy (Iverlewis procedure). Therefore the two-stage operation is the best option. Stage one chest cavity was cleaned, the lung decorticated, and thoracic duct ligated, and a thick pleural patch was wrapped around the perforated esophagus to limit the leak. A nasogastric tube was passed up to the 3rd part of the duodenum, and enteral feed was started. Once the patient was stable, ten days later as a second stage procedure was performed, and the feeding jejunostomy tube was fashioned to start postoperative enteral feeding. This is a very useful surgical technique in such cases. We managed our patient successfully, and he resumed back to his normal life.

Ethical approval

IRB approval.

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Author contribution

Ikram ul Haq Chaudhry, Main author operating surgeon, Abdullah M Al Ghamdi, Wrote abstract Assisting surgeon, Othman M Al Fraih, Wrote introduction Assisting surgeon, Hisham Al Maimon, Highlights, Yousif A Alqahtani, structured abstract, Farjad Tariq khan, Searched references, Fathi A AL Rasheed, Wrote part of discussion, Meenal A Al Abdulhai, Wrote part of discussion.

Registration of research studies

Name of the registry: Research registry.

Unique Identifying number or registration.

Hyperlink to your specific registration (must be publicly accessible and will be checked): http://www.researchregistry.com/browse-the-registry#home.

Guarantor

Ikram ul haq Chaudhry.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request"

Provenance and peer review

Not commissioned externally peer-reviewed.

Declaration of competing interest

No conflict of interest and there was no funding or financial assistance in this case.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.amsu.2022.103623.

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