CASE REPORT – OPEN ACCESS

International Journal of Surgery Case Reports 78 (2021) 197–200

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.casereports.com

Spontaneous perforation as a fatal presentation of esophageal tuberculosis: A case report

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ARTICLE INFO

Article history:
Received 27 November 2020
Received in revised form
11 December 2020
Accepted 15 December 2020

Keywords:
Esophageal tuberculosis
Perforation
Septic shock
Fatal case
Case report

ABSTRACT

INTRODUCTION: Tuberculosis is a multisystematic disease and is the most common cause of infectious disease–related mortality worldwide. Gastrointestinal tract is an uncommon site for extrapulmonary tuberculosis (TB). Esophageal TB is exceedingly rare.
PRESENTATION OF CASE: We report a 22-years-old male with esophageal TB that presented in septic shock from esophageal perforation. Despite all measures including surgical intervention and aggressive support in the intensive care unit, patient passed away.
DISCUSSION: The most common mechanism for esophageal involvement is secondary to direct spread from mediastinal structures and/or spreading the inoculation of swallowed sputum, or hematogenous or lymphatic spread. Once the diagnosis of TB is established, antibiosis is the cornerstone of treatment. Surgery is reserved only for complications of TB such as fistula, abscess, strictures or perforation. Less than 50% of cases are diagnosed within 24 h, and delay in diagnosis lead to significant increases in the mortality.
CONCLUSION: In countries with high prevalence of TB, this diagnosis should be considered in those with esophageal perforation with no underlying etiology and medical treatment for TB should be initiated in addition to conventional treatment in appropriate group of patients.
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1. Introduction

Tuberculosis has been one of the oldest diseases for centuries while threatening the life of human, It is currently estimated that over 2 billion people worldwide are exposed to mycobacterium tuberculosis, with over 8.7 million new infections in 2012, leading to 2.5–3 million cases of mortality per year; this makes tuberculosis the foremost cause of death by infection. One of the most important United Nation Millennium Development Goals is to control TB. About 85% and 15% of the TB cases are pulmonary and nonpulmonary, respectively [1,2]. Based on a report by WHO, in 2011, Iran had a lower incidence of tuberculosis (21 per 100,000 population) than all the neighboring countries (Turkey: 24 per 100,000, Iraq: 45 per 100,000, Armenia: 55 per 100,000, Azerbaijan: 113 per 100,000, Afghanistan: 189 per 100,000, and Pakistan: 231 per 100,000 [2]. Gastrointestinal (GI) tract mucosa is resistant to tuberculosis (TB) invasion and therefore alimentary tract is an uncommon site for extrapulmonary TB. Esophageal involvement by mycobacterium tuberculosis is very rare and less than 3% of the GI tract TBs are reported to involve this organ [3]. Primary infection of the esophagus is exceedingly rare, and involvement of this organ is commonly seen secondary to spread from adjacent mediastinal disease. Dysphagia is a common complaint among patients with esophageal TB and involvement of this organ can manifest with perforation or stricture [4]. Based on the **S**urgical **Ca**se **Re**port, 2018 (SCARE) guidelines [5], we present a case of Afghan young man with esophageal TB that presented with perforation.

2. Case report

A 22-year-old Afghan migrant worker male who were brought to our emergency room with dysphagia, chest pain, fever and, chills. He has a Body Mass Index (BMI) of 16. His symptoms started 7 days prior to presentation and worsening of his symptoms prompted an

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https://doi.org/10.1016/j.ijscr.2020.12.042
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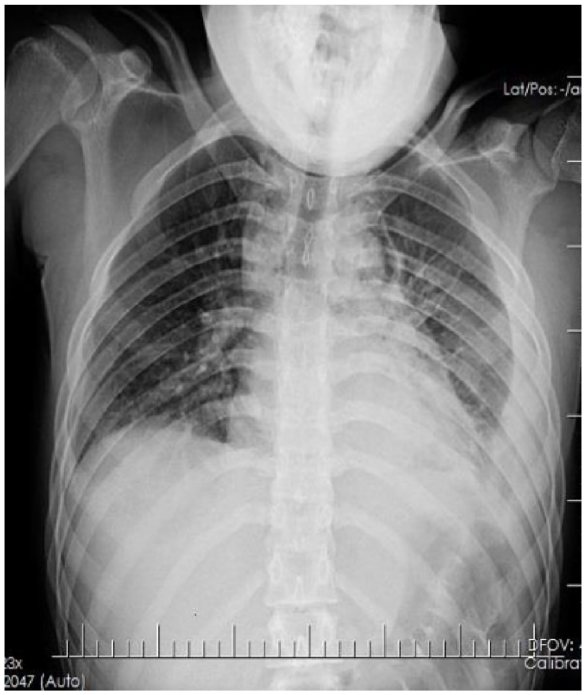

Fig. 1. Chest X-ray showing left side pleural effusion.

emergency room visit. His past medical and surgical history were unremarkable, and he did not report any recent upper endoscopy and he had a history of prolonged fever and weight loss from 6 months ago, as well as a history of dealing with the suspected TB patient. The patient drug and allergy history were negative. During the initial evaluation, he was found to be febrile, tachycardic (pulse rate: 130/min) and tachypneic (respiratory rate: 28/min), with oxygen saturation of 89%. Chest X-ray showed a left side effusion (Fig. 1) and a subsequent CT scan reconfirmed the effusion with a left side pneumothorax and pneumomediastinum (Fig. 2). Extraluminal contrast observed in the mediastinum was consistent with distal esophageal perforation. A chest tube was inserted in the left hemithorax with immediate return of 700 cc purulent fluid. We continued aggressive resuscitation and transferred the patient to the operating room for an emergent thoracotomy. Intraoperatively, he was found to have a large perforation in the distal esophagus, the decision was made to perform esophagectomy with cervical esophagostomy and gastrostomy.The procedure was performed by attendig thoracic surgeon.Patient was then transferred to surgical intensive care unit for continued resuscitation and ventilatory support. Despite all efforts, he remained in persistent septic shock and multi-organ failure and passed away on day 4 after surgery.

Detection of mycobacterial DNA by polymerase chain reaction (PCR) in the plural fluid and surgical pathology of the esophagus confirmed the diagnosis of mycobacterium tuberculosis (Fig. 3).

3. Discussion

Tuberculosis is a multisystemic disease and is the most common cause of infectious disease–related mortality worldwide. Studies report that this disease is becoming more common in many parts of the world [6]. Gastrointestinal (GI) tract is an uncommon site for extrapulmonary tuberculosis representing 1.2% of all TB cases [3]. Terminal ileum, cecum, and the peritoneum are the most common sites for GI and intraabdominal involvement of TB. Esophageal TB is a very rare condition, estimated to account for only 0.15% of deaths from tuberculosis. Previous studies have suggested that erect posture, esophageal peristalsis, stratified squamous epithelium, and saliva play a protective role against invasion of mycobacterium tuberculosis to esophageal mucosa [7]. In fact, many patients with esophageal TB have found to have underlying mucosal damage, such as Barrett's esophagus or esophageal cancer. Primary esophageal TB without active extraesophageal TB is exceedingly rare. The most common mechanism for esophageal involvement is secondary to direct spread from mediastinal structures, this explains the higher rate of tuberculosis in the middle-third of the esophagus which is located around the carina [8]. Other mechanisms for infecting esophagus include spreading the inoculation of swallowed sputum, or hematogenous or lymphatic spread.

The esophageal perforation in the patient in this case report was found to be in the distal esophagus, which is an uncommon site for TB involvement. It is possible that he had an underlying mucosal damage in the distal esophagus such as advanced esophagitis or Barrett's esophagus that increased the susceptibility of the esophageal lining to the invasion of mycobacterium tuber-

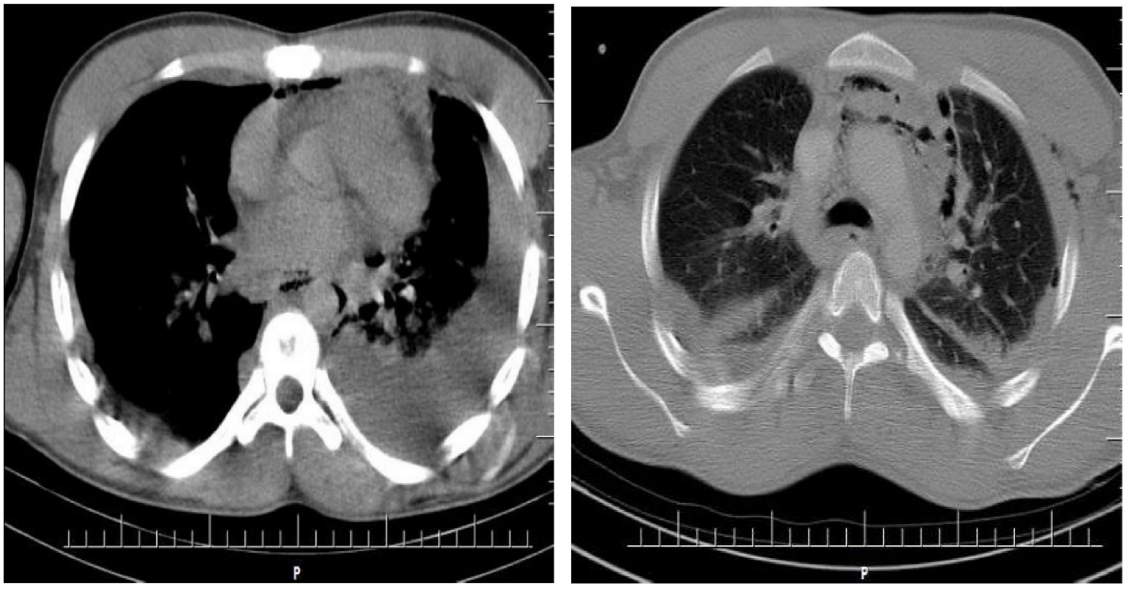

Fig. 2. Chest CT scan showing left side pleural effusion and pneumomediastinum.

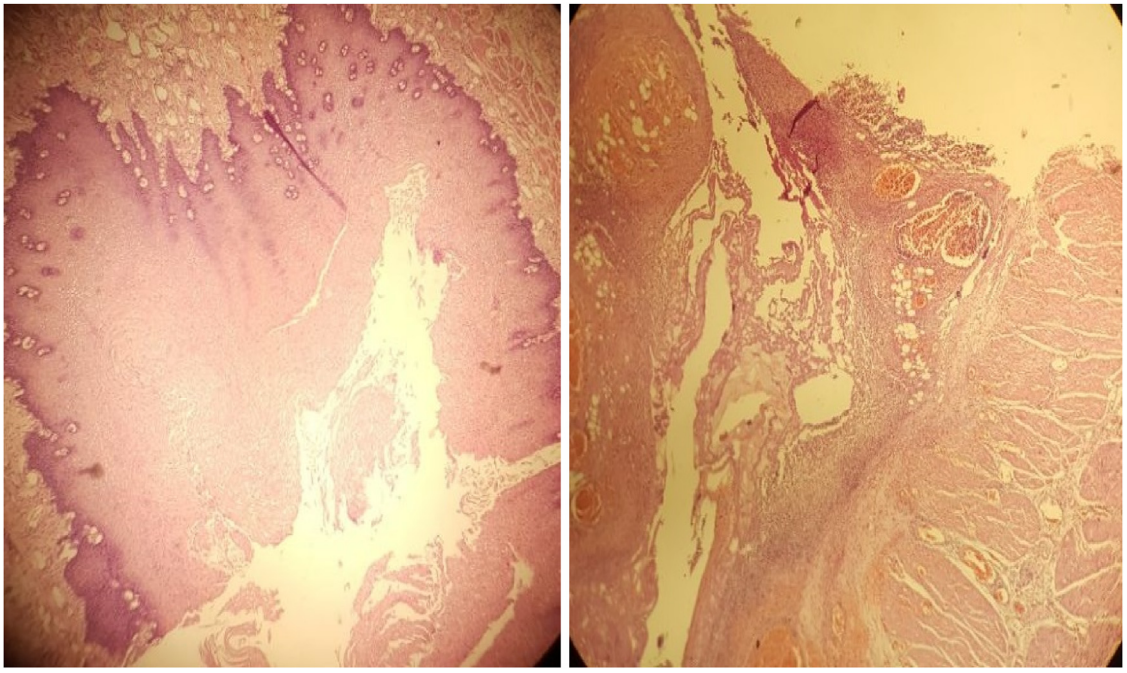

Fig. 3. Granuloma and exudative inflammation caused by TB.

culosis [9]. This hypothesis is hard to prove as our patient did not have any endoscopy prior to his fatal presentation to the emergency room.

In this patient we established the diagnosis of TB based on the results of polymerase chain reaction (PCR) analysis of the pleural fluid and histopathologic examination of esophagus. These two methods are the mainstay tests to confirm the diagnosis of TB. Recognizably, findings such as prolonged fever, shortness of breath, sweating, and weight loss should cast doubt on the condition. Once the diagnosis is established, antibiosis is the cornerstone of treatment. For initial empiric treatment of TB, patients are started on a 4-drug regimen: isoniazid, rifampin, pyrazinamide, and either ethambutol or streptomycin. Surgery is reserved only for complications of TB such as fistula, abscess, strictures or perforation [10]. Spontaneous esophageal perforation in the absence of a known underlying disease or excessive vomiting is rare. Less than 50% of cases are diagnosed within 24 h, and delay in diagnosis lead to significant increases in the mortality [4]. Our patient was symptomatic for 7 days and presented to us in septic shock.

In this case, presence of extraluminal oral contrast proved the diagnosis of esophageal perforation. Once this diagnosis is established, patient will require surgical or endoscopic intervention after initial resuscitation and antibiotic treatment. Surgical intervention should not be delayed achieving source control, otherwise profound sepsis and poor prognosis will be inevitable. Another choice for these patients could be thoracic lavage and drainage with esophagostomy and gastrostomy like a damage control surgery to minimize excessive surgical stress. In this patient, we decided to have a bigger operation due to the extent of the disease and the high pollution of the mediastinum.

In conclusion, esophageal perforation from tuberculosis is extremely rare, however in countries with high prevalence of TB, this diagnosis should be considered in those with esophageal perforation with no underlying etiology. Further, antibiosis for tuberculosis coverage should be initiated in appropriate group of patients with esophageal perforation with no response to conventional antibiotics and adequate surgical source control.

Declaration of Competing Interest

The authors report no declarations of interest.

Funding

None.

Ethical approval

This is a case report paper.

Consent

Informed consent was obtained from the legal guardian of the patient (his father), 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.

Author contribution

KH.A, M.S, J.Z.B, and SH.Q conceived and designed the study and wrote the manuscript; J.Z.B, SH.Q, S.M.F helped collect data; M.S wrote the manuscript; KH.A, S.M.F confirmed the eligibility of the participants' for the study; KH.A and M.S Supervised the whole study and approved the final version of the manuscript.

Registration of research studies

Not applicable.

Guarantor

Corresponding author is Dr. Javad Zebarjadi Bagherpour accept full responsibility for the work and/or the conduct of the study, had access to the data, and controlled the decision to publish.

Provenance and peer review

Not commissoned, externally peer-reviewed.

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