Case Report

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Nonhealing oesophageal ulcer: a case report

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Introduction and importance: Esophageal tuberculosis is a rare presentation of a common infectious disease. It may occur as a primary infection of the esophagus or as a secondary spread mostly from caseating mediastinal lymph nodes. The clinical diagnosis of the condition is presumed to be complex, owing to nonspecific biopsy findings, failure of isolation of bacilli, and a lack of predisposing conditions in patients. This study aims to present a rare condition of esophageal tuberculosis secondary to mediastinal lymphadenitis and highlights a unique modality of diagnosis of the condition, especially in a resource strained setting.

Case presentation: This case report presents the case of a 50-year-old male with dysphagia and a burning sensation at the epigastrium. Endoscopy and histopathological examination showed ulceration at the esophagus and granulomatous inflammation, respectively. Computed tomography showed enlargement of the prevascular and paratracheal group of lymph nodes. However, the acid-fast bacilli stain at the ulcer site was negative. The diagnosis could be confirmed only after 2 months of the antitubercular treatment trial, which significantly potentiated ulcer healing.

Clinical discussion: Esophageal tuberculosis may result from a secondary infection caused by systemic dissemination following a pulmonary disease or as a primary infection. In this case, it likely resulted from lymphatic dissemination via prevascular and paratracheal lymph nodes manifested mainly as dysphagia.

Conclusion: Tuberculosis should be considered as one of the differential diagnoses in areas of limited resources. Clinicians may have to rely on clinical judgement and/or the patient's response to standard antitubercular treatment to make a definitive diagnosis.

Keywords: case report, dysphagia, endoscopy, esophagus, tuberculosis

Introduction

Esophageal tuberculosis is a rare entity, accounting for only 2.8% of all gastrointestinal tuberculosis[1]. Esophageal protective mechanisms like stratified squamous epithelium, saliva, peristalsis, and rapid transit make primary esophageal tuberculosis quite uncommon[2,3]. Secondary esophageal tuberculosis is a commoner phenomenon with direct extension from caseating mediastinal lymph nodes being the most common source of spread[4]. The most common site of involvement is the middle third of the esophagus around the carina[5]. Presenting symptoms include dysphagia, retrosternal pain, weight loss, and fever[5]. The clinical diagnosis of the condition is presumed to be complex, owing to nonspecific biopsy findings, failure of isolation of bacilli, and a lack of predisposing conditions in patients[4,6]. The presentation of the clinical and other relevant details of this case will facilitate providing evidence on its rare existence and additional information. This will direct future researchers to incorporate the limitations and generate more evidence so that this rare condition gets recognized. Thus, this case study aims to present a rare condition of esophageal tuberculosis secondary to mediastinal lymphadenitis evidenced by enlargement of prevascular and paratracheal lymph nodes. The case report was written following the CARE 2020 criteria[7].

HIGHLIGHTS

- The esophagus is usually protected by its stratified squamous epithelium and other mechanisms and tuberculosis of the esophagus is relatively rare.
- Diagnostic modalities are variable and mainly rely on endoscopy but were not confirmative.
- The diagnosis is confirmed mainly via a clinical suspicion and on the response to standard antitubercular therapy.
- The condition must be primarily distinguished from a carcinoma and a fungal infection of the esophagus.
- The ulcer responded to a standard course of antitubercular therapy.

Case report

A 50-year-old male presented to the outpatient clinic of our hospital, a tertiary-level care center in Nepal, with complaints of dysphagia and a burning sensation in the epigastric region for

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~15 days. The dysphagia was exacerbated mostly by swallowing food, and only to some extent while drinking fluids and swallowing saliva. He also had a fever with an evening rise for 2 weeks, associated with chills and sweating. During the fever, the maximum temperature recorded was 102 Fahrenheit. Within a span of two months, he lost about 5 kg of his body weight. Halitosis and a dry cough were also reportedly present one month before the onset of odynophagia, without any symptoms of hemoptysis.

The indexed case did not report any comorbidities. He reported that he was a past smoker and had quit a year ago. More importantly, some of his family members had also been diagnosed with tuberculosis. His son, 27, had suffered from pulmonary tuberculosis a year ago, and after undergoing regular antitubercular treatment (ATT), he has improved and recovered. Additionally, his daughter, 25, had also been diagnosed with pulmonary tuberculosis a month ago and is going through regular ATT. His clinical examination was normal and unremarkable. There was no significant drug history.

On his first visit to the outpatient clinic, an upper gastrointestinal endoscopy showed a deep longitudinal esophageal ulceration at about 26 cm from the lower incisors (Fig. 1). A biopsy of the site showed no evidence of malignancy. He was prescribed oral lansoprazole tablets and sent home. A follow-up report of the endoscopy after 3 weeks demonstrated a single cratered linear ulcer, 20 mm in diameter, without any elevated border or centripetal tear, seen at 28 cm from the lower incisors. A contrast-enhanced computed tomography of the chest and abdomen showed a few centimeter size prevascular and paratracheal lymph nodes, with the largest measuring 15 × 18 mm in the mediastinum. The hematology report showed decreased platelet count of 122 × 10^3/microL. The lung function test, creatinine, erythrocyte sedimentation rate, and C-reactive protein were within normal limits.

A histopathological examination conducted a month later showed granulomatous inflammation at the ulcer site. However, the acid-fast bacilli (AFB) stain was negative. In view of the lack of significant healing and a biopsy suggestive of granulomatous inflammation, an empirical ATT trial was started with four tablets of HRZE, that is, Isoniazid, Rifampin, Pyrazinamide, and Ethambutol, once a day for 2 months, and he was also prescribed other medicines such as oral pyridoxine and lansoprazole.

A repeat endoscopy was done a month later, which showed that nearly two-thirds of the ulcer had healed (Fig. 2). This confirmed the diagnosis of esophageal TB, and therefore, he was started on a full-fledged course of ATT. Six months later, a repeat endoscopy was done to check the recovery of the ulcer, which showed that it had healed completely, leaving some depressed areas (Fig. 3). The patient reports no significant adverse effect to the medications and has reported adherence to the intervention.

Discussion

Tuberculosis is a disease caused by the infection of bacteria belonging to the Mycobacterium tuberculosis complex and the related Mycobacterium avium complex[8]. Being a transmissible disease, the respiratory droplets containing the bacteria from infectious patients are inhaled by the susceptible hosts and reach their alveoli[8]. The bacilli may also gain systemic access via the blood and may involve extrapulmonary tissues during active infection, or they may remain latent and reactivate later in life depending on the levels of the host's immunity[9,10]. Esophageal tuberculosis may result from a secondary infection caused by systemic dissemination following a pulmonary infection as described above, or from a primary infection[3,11]. Clinically, dysphagia, odynophagia, chest pain, and nonspecific systemic symptoms such as fever, loss of appetite, and anorexia may also be commonly observed, with hematemesis being reported in severe cases[3]. The family members (son and daughter) of the indexed case were also reported as active tuberculosis cases during his presentation.

Diagnostic modalities involve upper gastrointestinal endoscopy and histology. Gross endoscopic findings commonly reveal lesions that are either ulcerative, granular, or hypertrophic[3,11]. On histology, it is characterized by Langerhans cells surrounding an area of central necrosis and the presence of AFB[11]. Radiological modalities reveal nonspecific findings consistent with the ulceration, strictures, and fistula. The case presented in this study reported mediastinal lymphadenitis[4]. Although such modalities do exist, results are often variable, similar to the case in this study, where the lesion showed no AFB, so the diagnosis was

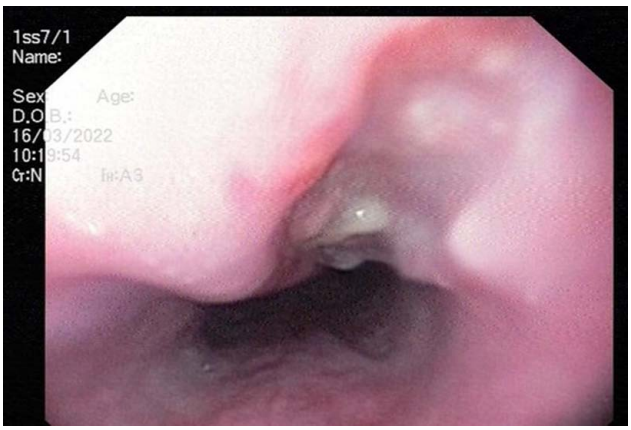

Figure 1. Endoscopy image showing longitudinal esophageal ulceration at about 26 cm from the lower incisors.

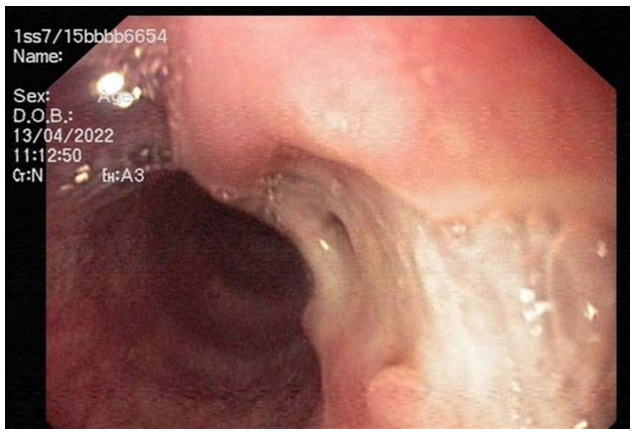

Figure 2. Repeat endoscopy image after about a month, showing healing of the majority of the ulcer after the inception of antitubercular therapy.

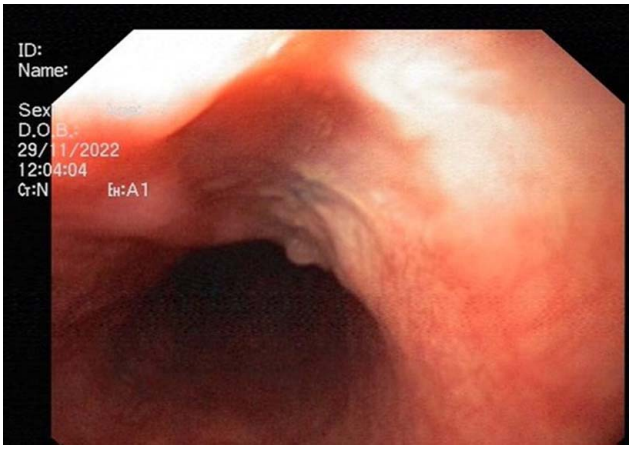

Figure 3. Follow-up endoscopy image after the completion of the antitubercular therapy of the ulcer showing complete healing with only a slightly depressed area.

based on clinical judgement. And importantly, this must be distinguished from conditions such as carcinoma and fungal infection of the esophagus as they may have a similar presentation[3,4].

The treatment of esophageal tuberculosis involves the usual antitubercular regimen[12]. The suggested dosage includes isoniazid, streptomycin, and rifampin with a satisfactory clinical response in 2–4 weeks, or ethambutol[4]. The treatment involves an intensive phase, which uses isoniazid, rifampicin, pyrazinamide, and ethambutol for a period of 2 months as an intensive phase, with a continuation phase of 4 months with isoniazid and rifampicin[12]. As mentioned above, we could establish a definite diagnosis only after observing treatment outcomes with the antitubercular regimen and endoscopic examination of the change in ulcer size. The case in this study started the medication with four tablets of HRZE, that is, Isoniazid, Rifampin, Pyrazinamide, and Ethambutol, and other medicines such as oral pyridoxine and lansoprazole. After a full-fledged course of the ATT, the case recovered, as confirmed by a repeat endoscopy procedure.

A major strength of this case study is that it reports a rare case that is difficult to diagnose, more so in resource-constrained settings. This case study has been reported despite several limitations. Future researchers are encouraged to report a few more cases to make the information on this particular disease condition more comprehensive. This case also highlights the limited modalities of diagnosis of esophageal tuberculosis, thus, future studies should consider other modalities as well to have a broad overview of the management of such cases.

Conclusion

Esophageal tuberculosis must be distinguished from conditions such as carcinoma and fungal infections. The presentation of esophageal tuberculosis may mimic a fungal infection or malignancy in terms of its appearance. In resource-constrained settings, tuberculosis should be considered as one of the differential diagnoses despite difficulties in its precise diagnosis. Variable results of endoscopy and histology may compel physicians to make a diagnosis based on their clinical judgement and/or the patient's response to standard ATT.

Ethical approval

None declared.

Consent

Written informed consent was obtained from the patient for the 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.

Consent

An informed consent was taken from the patient.

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Author contribution

R.B.G., P.Sapkota, and P.Sharma: endoscopic procedure, patient care, manuscript review; A.L. and S.J.: manuscript writing and editing; U.P., T.P.P., and S.B.: manuscript editing.

Conflicts of interest disclosure

All the authors declare that they have no conflicting interests.

Research registration unique identifying number (UIN)

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