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Single Case

Esophageal Tuberculosis as a Rare Cause of Dysphagia: Case Report

Billy Zhao[a] Hyun Jae Kim[b] Jessica Farrell[c] Wei Xiong[d] Jennifer Telford[e] Sarvee Moosavi[f]

[a]Department of Medicine, University of British Columbia, Vancouver, BC, Canada; [b]Division of Gastroenterology, University of British Columbia, Vancouver, BC, Canada; [c]Department of Radiology, Mount St. Joseph Hospital, Vancouver, BC, Canada; [d]Department of Pathology, University of British Columbia, Vancouver, BC, Canada; [e]Division of Gastroenterology, St. Paul's Hospital, University of British Columbia, Vancouver, BC, Canada; [f]Division of Gastroenterology, Vancouver General Hospital, University of British Columbia, Vancouver, BC, Canada

Keywords
Dysphagia · Tuberculosis · Esophagus · Submucosal mass

Abstract
Introduction: The esophagus and duodenum are rare sites of manifestation for extrapulmonary tuberculosis (TB). Its rarity makes the diagnosis challenging, especially when no other organ is involved, and the endoscopic findings may resemble malignancy. ***Case Presentation:*** We report a unique case of a 37-year-old woman who presented with dysphagia secondary to esophageal TB with an endoscopic appearance of a submucosal mass resembling malignancy. ***Conclusion:*** Esophageal TB is a rare cause of dysphagia, especially in a western setting. It should always be considered as a potential etiology in patients with dysphagia.

Introduction

Gastrointestinal tuberculosis (TB) is a rare extrapulmonary manifestation of TB [1]. Involvement of terminal ileum, cecum, and peritoneum are more commonly documented while esophageal involvement accounts for less than 0.2% of all TB patients [2–4]. Despite the lower incidence of TB in Western countries, given the vague clinical manifestations of gastrointestinal TB, clinicians are required to keep TB on their differentials in work-up of various gastrointestinal symptoms. We present a unique case of esophageal and duodenal TB in a patient with a prior negative Mantoux tuberculin skin test (TST).

Correspondence to:
Billy Zhao, billy.zm.zhao@gmail.com

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Case Report

The CARE Checklist has been completed by the authors of this case report and is attached as online supplementary material (for all online suppl. material, see https://doi.org/10.1159/000540292). A 37-year-old female from the Philippines presented to the emergency department with a 4-month history of progressive dysphagia to solids and then liquids. She was otherwise healthy and did not take any prescribed medication. She denied odynophagia, food bolus obstruction, chest pain, regurgitation, heartburn, and respiratory symptoms. She had unintentional weight loss of 3 kg over 4 months with no other constitutional symptoms.

The patient had received Bacillus Calmette-Guérin (BCG) vaccination in the Philippines as a child. Her chest X-ray on Canadian immigration screening in 2009 was negative for latent or active TB. Her only known TB exposure occurred 14 months ago when her mother visited Canada and was diagnosed with active pulmonary TB after returning to the Philippines. Interestingly, she had a negative TST as a part of her care aide employment screening 5 months before her emergency department presentation.

Cardiac, respiratory, and abdominal examinations revealed no abnormalities with no palpable lymphadenopathy. Complete blood count with differential showed mild normocytic anemia with hemoglobin of 108 g/L (115–155). WBC was normal at 5.3×10^9/L (4.0–11.0). She had a normal neutrophil count of 3.6×10^9/L (2.0–8.0), mildly decreased lymphocyte at 1.1×10^9/L (1.2–3.5), and normal monocyte, eosinophil, and basophil counts. Electrolytes and hepatic panels were normal. Her chest X-ray in the emergency department showed no signs of lung scarring.

Esophagogastroduodenoscopy (EGD) revealed a 2 cm ulcerated mass, abutting the gastroesophageal junction (GEJ) inferiorly on retroflexion (Fig. 1) and a subepithelial lesion in the duodenal bulb with normal overlying mucosa. The exam was otherwise normal. Mucosal biopsies were taken from both lesions and histopathology revealed ulcerated mucosa with acute and chronic inflammatory infiltrates composed of histiocytes, granulocytes, and lymphocytes (Fig. 2). No granulomas were identified. Ziehl-Neelsen stain was negative for acid-fast bacilli. Although a lymphoproliferative disorder was not identified, it could not be excluded in the limited tissue sample.

Additionally, computed tomography (CT) scan of her chest, abdomen, and pelvis was performed for staging due to suspicion of lymphoma or other malignancy. This demonstrated infiltrative soft tissue masses at the GEJ and duodenal bulb, each measuring up to 4.5 cm in size (Fig. 3). There was linear scarring and several discrete nodules at the left lung apex, the largest 12 mm (Fig. 4). No pathologically enlarged lymph nodes were noted.

Endoscopic ultrasound (EUS) demonstrated an ill-defined, hypoechoic lesion, involving the GEJ, extending into the region of the celiac axis. A second lesion, involving the proximal duodenum, was round with well-defined margins and measured 20 mm × 14 mm (Fig. 5). No enlarged lymph node was seen. EUS-guided fine-needle biopsy of both lesions was performed, which revealed fragments of necrotizing granulomatous inflammation (Fig. 6). No mycobacteria were seen on Ziehl-Neelsen stain (Fig. 6). Concurrent flow cytometry was completed with a 10-color lymphoma screening panel. The CD45 gated lymphocyte population accounted for 4% of all events. T-cells were 66% of the lymphocyte gate and showed normal antigen expression. The CD4/CD8 ratio is 1.7. Overall, the flow cytometry analysis was not consistent with lymphoproliferative disorders. Given this finding along with the fact that previous infectious investigations are not highly sensitive, infectious etiologies cannot be ruled out, and the patient was referred to the infectious disease team and underwent an IGRA/QuantiFERON assay, which came back positive. Diagnosis of TB was further confirmed by a positive TB PCR testing on paraffin block on esophageal biopsies from index EGD. The patient was subsequently referred to a TB-specialized clinic and was started on Rifampin,

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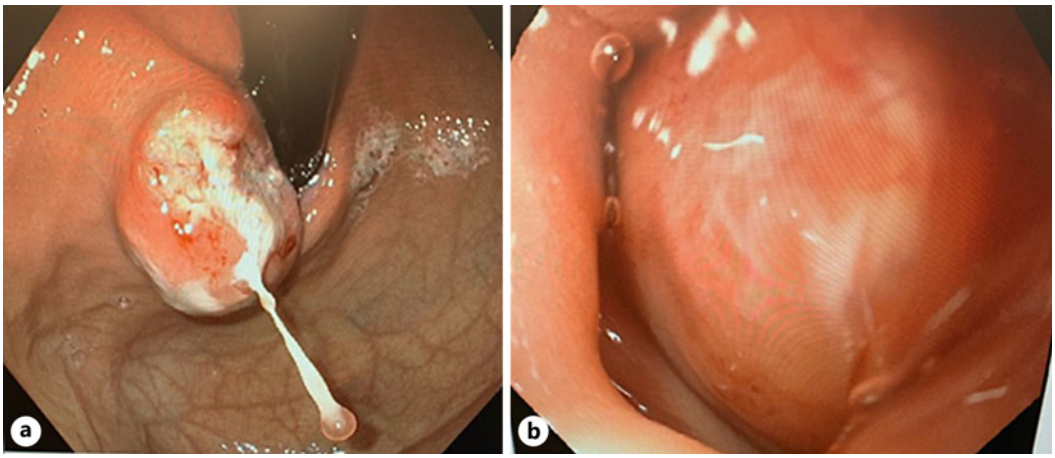

Fig. 1. Esophagogastroduodenoscopy showing an ulcerated, bulging 20 mm mass abutting the gastroesophageal junction on retroflexion (**a**) and submucosal mass at the proximal duodenal bulb (**b**).

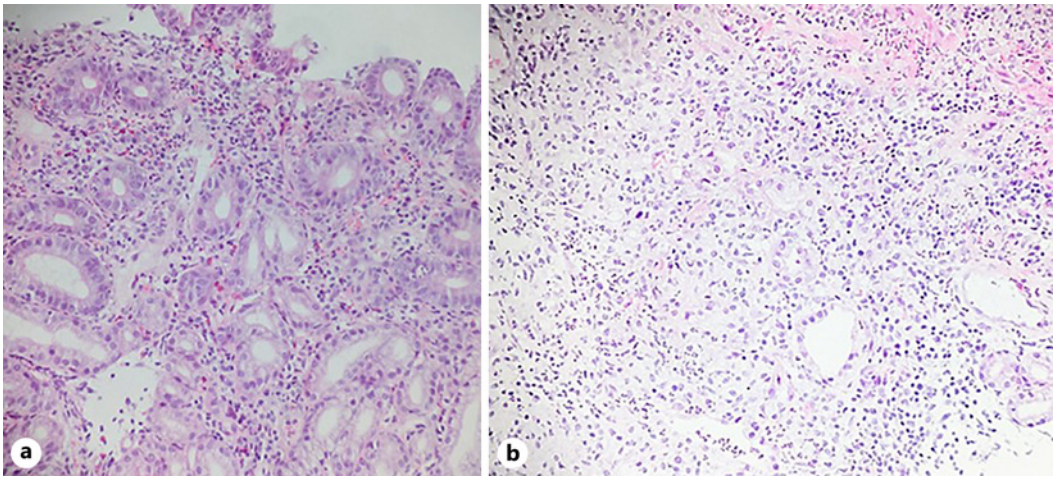

Fig. 2. Biopsy from index esophagogastroduodenoscopy showing active inflammation and reactive epithelial changes on gastroesophageal junction mass (**a**; H&E stain, ×200) and inflammation with histiocytes and neutrophils on duodenal submucosal mass (**b**; H&E stain, ×100).

isoniazid, vitamin B6, and pyrazinamide for 6 months. Given the lack of respiratory symptoms or active findings on chest imaging, isolation was not required, and her immediate family members were all screened for TB.

Discussion

Dysphagia is the most common presenting symptom of esophageal TB; however, this is a common presenting symptom of a rare disease [5–7]. Other symptoms of esophageal TB include chest pain, fever, odynophagia, and weight loss.

Endoscopic findings of esophageal TB can vary, but typically present as either ulcerated, bulging mucosal, or submucosal mass and rarely as a stricture or fistula [5, 8–10]. Given its

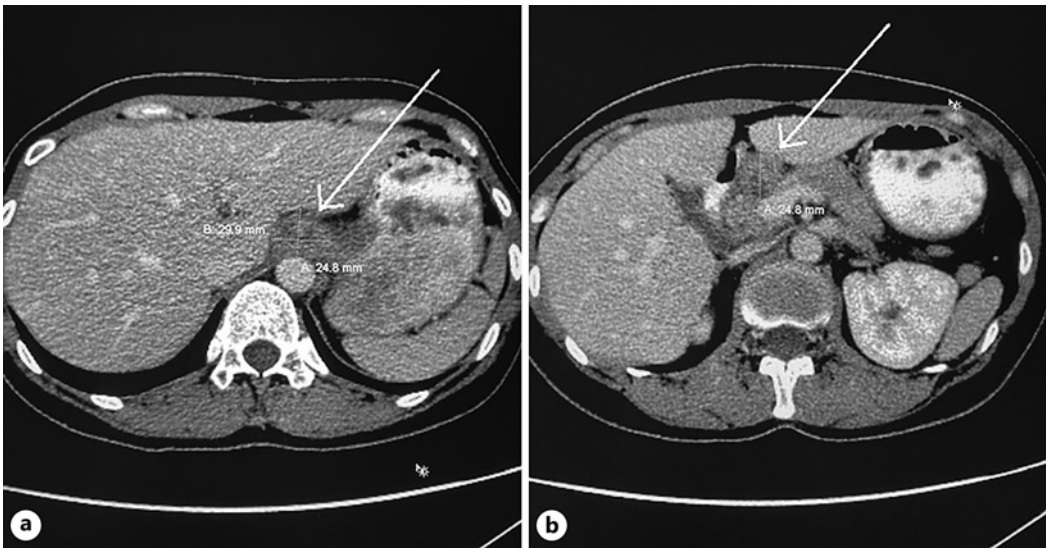

Fig. 3. Axial view of CT abdomen with oral contrast and IV contrast in portal venous phase showing the gastroesophageal junction mass (**a**) and the submucosal mass at the pyloric channel opening (**b**).

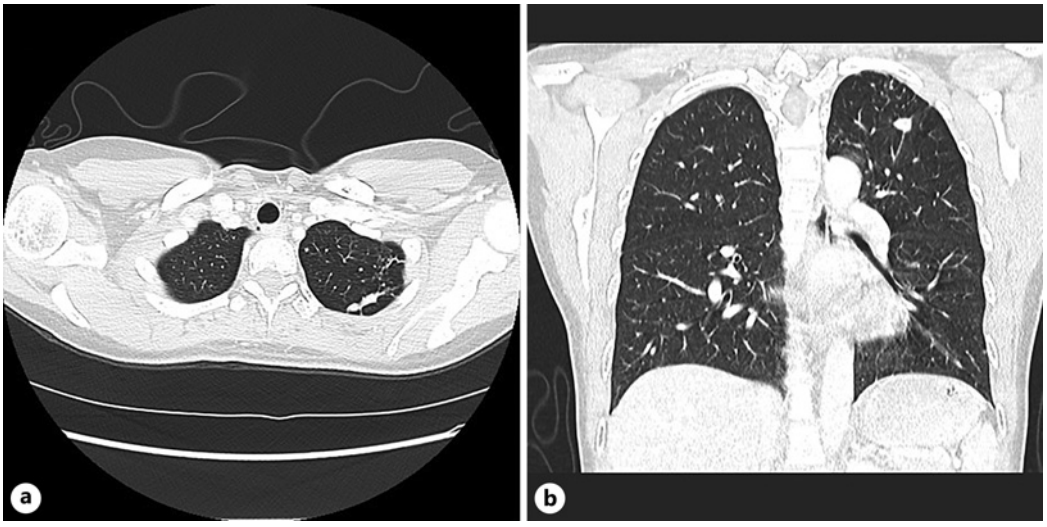

Fig. 4. Axial (**a**) and coronal (**b**) view of CT chest with IV contrast showing a nodular band-like opacity 6 mm in thickness at the left lung apex (**a**), with adjacent discrete nodules, the largest at 12 mm in size (**b**). Associated linear scarring, focal mild bronchiolectasis, and volume loss were suggestive of post-infectious scarring. No calcified pulmonary nodule was identified.

appearance, upper gastrointestinal TB is often mistaken for malignancy in reported cases, as it was in our case [11, 12].

Adding to the diagnostic dilemma was the fact that our patient was both previously vaccinated with BCG and had a prior negative TST 5 months before presentation. BCG has an effectiveness of 46–50% regardless of the time of vaccination [13, 14]. TST has a sensitivity of 60–98% but is prone to false-negative results from technical errors or decreased skin reactivity [15]. Acid-fast bacilli staining on biopsied tissue has high specificity but low sensitivity for TB [16]. Instead, tissue PCR should be used, given the higher sensitivity estimated

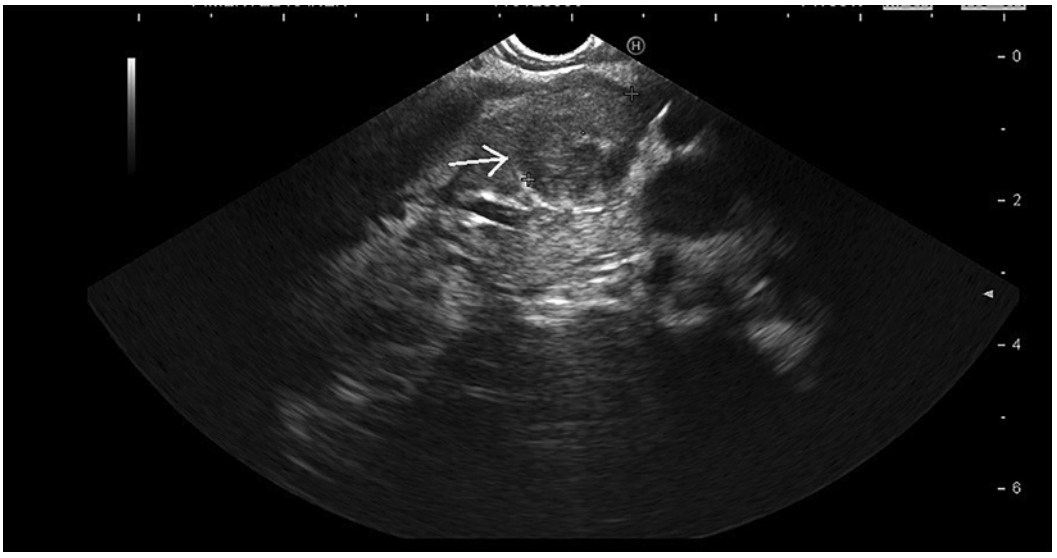

Fig. 5. Endoscopic ultrasound image of the submucosal lesion at the duodenal bulb, just distal to pyloric channel.

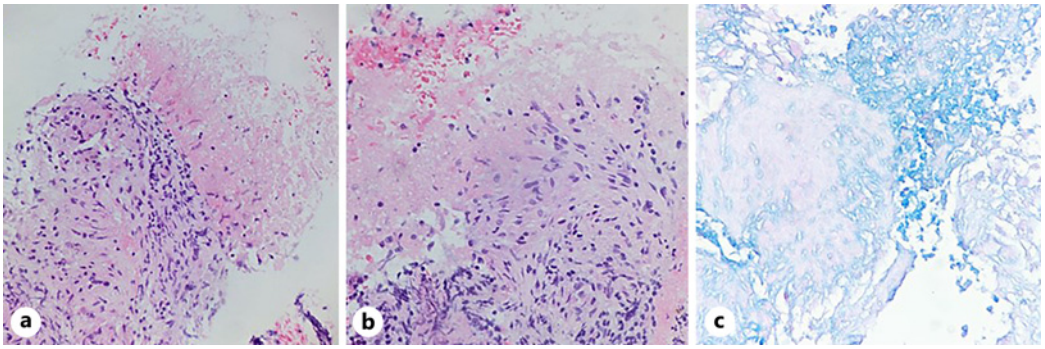

Fig. 6. Endoscopic ultrasound guided fine-needle aspiration showing necrotizing granuloma at gastroesophageal junction mass (**a**; H&E stain, ×200) and duodenal bulb (**b**; H&E stain, ×200) with Ziehl-Neelsen stain negative for mycobacteria (**c**; ×400).

to be between 74% and 100% with most false-negatives as a result of insufficient tissue sampling [16–18].

The most common etiology of esophageal TB is local spread from adjacent respiratory tract or mediastinal lymph nodes [5, 19]. While our patient had no respiratory symptoms, normal chest X-ray, and negative TST, lung scarring seen on her CT chest may indicate latent pulmonary TB. Lymphadenopathy was absent on both EUS and cross-sectional imaging, making spread from lymph nodes less likely.

Other than enlarged lymph nodes, other EUS features of esophageal TB include thickened esophageal wall and hypoechoic lesions that may contain hyperechoic calcifications, which we did not see in our patient [19]. However, none of these features is sensitive nor specific to esophageal TB. Primary esophageal TB is also unlikely as it is rare and occurs when mycobacterium TB adheres to the esophageal wall after direct ingestion and needs to bypass protective mechanisms such as the mucus barrier and stratified squamous epithelium of the esophagus [9, 19].

Despite previous vaccination, a negative skin test, and the absence of other features of TB other than generalized weight loss, our patient was found to have esophageal and duodenal TB. Although rare in a Western setting, an index of suspicion is needed to keep gastrointestinal TB as part of the differential for work-up of dysphagia for patients from endemic areas, even with prior BCG vaccination and negative screening TST.

Statement of Ethics

Ethical approval is not required for this study in accordance with local or national guidelines. Written informed consent was obtained from the patient for publication of the details of their medical case and any accompanying images.

Conflict of Interest Statement

None of the authors have any conflict of interest to declare.

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Author Contributions

Conception and design: Billy Zhao, Sarvee Moosavi. Data collection, interpretation, and analysis: Billy Zhao, Hyun Jae Kim, Jessical Farrell, Wei Xiong, Jennifer Telford, and Sarvee Moosavi. Drafting of the article: Billy Zhao. Critical revision of the article for important intellectual content: Hyun Jae Kim, Jessical Farrell, Wei Xiong, Jennifer Telford, and Sarvee Moosavi. Final approval and guarantor of the article: Sarvee Moosavi.

Data Availability Statement

All data generated or analyzed during this study are included in this article and its online supplementary material. Further inquiries can be directed to the corresponding author.

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