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Case Report
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An Unusual Case of Dysphagia in an Adolescent Boy

Cierra W[1], Christine D[2], and Dhiren P[2,*]

[1]Department of Pediatrics, Saint Louis University, Cardinal Glennon Children's Hospital, Saint Louis, Missouri, USA
[2]Pediatric Gastroenterology, Saint Louis University, Cardinal Glennon Children's Hospital, Saint Louis, Missouri, USA

[*]**Corresponding author:** Dhiren P, Pediatric Gastroenterology, Saint Louis University, Cardinal Glennon Children's Hospital, Saint Louis, Missouri, USA; E-mail: dr_dhiren_patel@yahoo.com

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Abstract

This case is a rare presentation of tularemia. One other case has been reported in the literature describing tularemia in an otherwise healthy adult with pulmonary infiltrates, as well as mediastinal and hilar necrotizing lymphadenopathy. This case demonstrates the importance of the differential diagnosis when evaluating patients with dysphagia and/or odynophagia. Extrinsic compression of the esophagus should be considered and evaluated for carefully particularly if endoscopy is suggestive.

Keywords: Dysphagia; Thoracoscopic; *Francisella tularensis*

Case Presentation

A 13 year old male presented with a 10-day history of progressively worsening epigastric pain with acute onset of dysphagia and odynophagia with solids. He endorsed periodic chest tightness and sensation of food becoming stuck in his upper chest upon swallowing, but denied vomiting, decreased appetite, weight loss, fever, diarrhea, or bloody stools. He did not have a history of anxiety disorder, cardiac disease, or pulmonary disease. Of note he was diagnosed clinically with Lyme disease after a tick bite to the left arm one and half months prior to presentation. He was treated with doxycycline for 2 days, however, this was discontinued secondary to abdominal pain. He then completed a two week course of amoxicillin-clavulanic acid. In addition, four days prior to presentation, the patient sustained a left elbow fracture and arm was casted covering the initial site of the tick bite [1-3].

Upon admission, he underwent evaluation with an esophagogastroduodenoscopy which revealed a tortuous and irregular esophageal course suggesting possible extrinsic compression of the mid-esophagus with erythema of the overlying mucosa (Figure 1). Histology showed only mild esophagitis without significant eosinophilic infiltration. An esophagram was performed showing extrinsic compression of the mid-thoracic esophagus with no obstruction (Figure 2). A magnetic resonance imaging of the chest with and without contrast demonstrated a 4.8 cm heterogeneously enhancing solid subcarinal mass, hilar lymphadenopathy, and right-sided pulmonary nodules (Figure 3). The patient underwent thoracoscopic biopsy and resection of the posterior mediastinal mass. Histological assessment demonstrated necrotizing granuloma formation with fibrosis and calcification, however, no acid-fast bacilli, yeast, or hyphae were observed.

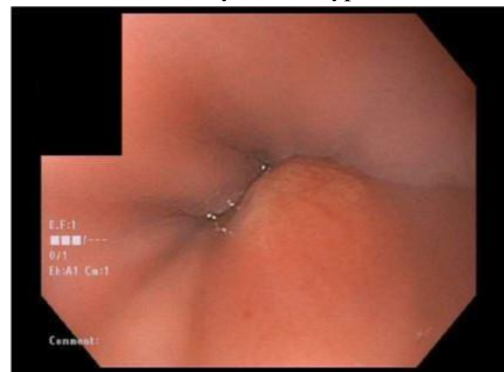

Figure 1: *Mid-esophagus with erythema of the overlying mucosa.*

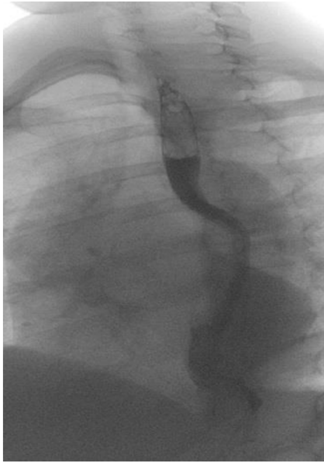

Figure 2: *Mid-thoracic esophagus with no obstruction.*

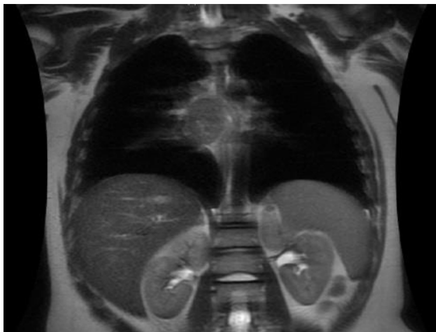

Figure 3: *Hilar lymphadenopathy, and right-sided pulmonary nodules.*

Due to concern for granulomatous disease, infectious disease was consulted and additional testing was negative for histoplasmosis and lyme disease. In outpatient follow-up, the family noted that upon removal of the patient's cast on his left arm, he had purulent drainage from the site of the previous tick bite. He was subsequently tested at this visit for tularemia and serum IgG for *Francisella tularensis* was found to be significantly elevated with a titer of 1:64 and IgM was negative. He was given an empiric course of ciprofloxacin at the time of lab draw.

Discussion

The differential diagnosis for pediatric patients with dysphagia includes etiologies such as eosinophilic esophagitis, infectious esophagitis, anatomical abnormalities, achalasia/esophageal motility disorders, behavioral causes, malignancy, and extrinsic compression. This case represents an unusual etiology of dysphagia in a previously healthy adolescent male. After careful endoscopic evaluation and imaging, it was obvious that mucosal disease was not present. Extrinsic compression of the esophagus can be a result of infectious, anatomical, or oncological etiologies. In our case, this compression was due to necrotizing granuloma from a tularemia infection.

Francisella tularensis is a gram-negative coccobacillus which can be transmitted to humans via tick bite or through the handling of infected animals. Symptoms vary depending on mode of bacterial entry. Main forms of tularemia include ulceroglandular, glandular, oculoglandular, oropharyngeal, pneumonic, and typhoidal. Treatment includes appropriate antibiotic selection and course. Common choices include streptomycin, gentamycin, fluoroquinolone, or tetracycline. Our patient was treated with ciprofloxacin 500 mg twice daily for a 7 day course.

Conclusion

This case is a rare presentation of tularemia. One other case has been reported in the literature describing tularemia in an otherwise healthy adult with pulmonary infiltrates, as well as mediastinal and hilar necrotizing lymphadenopathy. This case demonstrates the importance of the differential diagnosis when evaluating patients with dysphagia and/or odynophagia. Extrinsic compression of the esophagus should be considered and evaluated for carefully particularly if endoscopy is suggestive.

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