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Esophagomediastinal fistula due to secondary esophageal tuberculosis: report of two cases

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Abstract

Esophageal tuberculosis is rare among digestive system diseases. We herein present two cases of esophageal tuberculosis. One patient presented with a choking sensation and pain in the chest, and the other presented with loss of appetite and emaciation. Both patients had an esophagomediastinal fistula, underwent endoscopic ultrasonography and fine-needle aspiration, were clinically diagnosed with esophageal tuberculosis, received antituberculosis treatment, and exhibited clinical improvement. These two rare cases suggest that the possibility of esophageal tuberculosis should be considered in patients with an esophagomediastinal fistula. Endoscopic ultrasonography and fine-needle aspiration can be performed to assist the diagnosis. Good clinical results can often be achieved with timely antituberculosis treatment.

Keywords

Esophageal tuberculosis, esophagomediastinal fistula, endoscopic ultrasonography, fine-needle aspiration, granulation tissue, antituberculosis treatment

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Introduction

Esophageal tuberculosis is a granulomatous inflammatory disease of the esophageal wall caused by *Mycobacterium tuberculosis* infection. It is rare among digestive system diseases, accounting for about 0.20% to

1.00% of all cases of gastrointestinal tuberculosis.[1–5] We herein present two cases of esophageal tuberculosis.

Case presentation

Patient 1

A 68-year-old man presented with distention and pain in the chest. At presentation, he had a choking sensation when eating. The patient's purified protein derivative (PPD test) and antituberculosis antibody test were positive, and his T-cell immunospot test (T-SPOT) was strongly positive (120 spot-forming cells [SCFs] per 2.5×10^5 peripheral blood mononuclear cells [PBMCs]). Computed tomography revealed thickening of the middle esophageal wall and multiple enlarged lymph nodes in the hilum and mediastinum, some with calcification and some with an unclear boundary between the lymph nodes and middle esophagus (Figure 1(a)). After admission, the patient underwent endoscopic ultrasonography and fine-needle aspiration (EUS-FNA). Under endoscopy, we found a 1.0- × 2.5-cm longitudinal fistula in the esophageal anterior wall located 27 to 29 cm from the incisor (Figure 1(b)). Pus overflow was observed from the fistula, and no bubbles were seen. Ultrasound examination showed that the tissue layers outside the esophageal wall were unclear, and several of the multiple lymph nodes under the carina and around the esophagus were calcified (Figure 1(c)). Six biopsies were performed at the base of the fistula, which exhibited hard tissue and poor elasticity. The lymph nodes were then punctured four times with a fine needle (Figure 1(d)), and tissue strips and black carbon foam were observed. Microscopically, the biopsy showed active chronic inflammation, granulation tissue formation, local necrosis (Figure 1(e)), negative acid fast staining, and negative tuberculosis-polymerase chain reaction (TB-PCR). The tissue from the lymph node puncture showed inflammatory exudates, lymphoid cells, and tissue cells that phagocytized carbon foam. Based on these clinical manifestations, the patient was clinically diagnosed with esophageal tuberculosis and underwent standard antituberculosis treatment after providing informed consent. His symptoms then gradually decreased in severity, and the fistula had healed when he was re-examined by endoscopy 6 months later (Figure 1(f)).

Patient 2

A 70-year-old woman presented with loss of appetite and emaciation. Her PPD test was strongly positive, and her T-SPOT was positive (22 SCFs per 2.5×10^5 PBMCs). Positron emission tomography–computed tomography revealed an esophagomediastinal fistula and multiple enlarged lymph nodes in the hilum and mediastinum (Figure 2(a)). After admission, the patient also underwent EUS-FNA. Under endoscopy, we found a 0.5- × 0.4-cm deep ulcer-like lesion in the esophageal anterior wall located 26 cm from the incisor (Figure 2(b)). Granulation tissue could be seen on the oral side, and the surface of the ulcer was covered with white pus. Ultrasound examination showed an 1.8- × 1.5-cm hypoechoic mass lesion beside the esophageal ulcer; the internal echo of the lesion was not uniform, and fusion of lesions was seen within it. A sinus could be seen between the esophageal lesion and the mass (Figure 2(c)). Four biopsies were performed at the base of the ulcer, which exhibited firm tissue and fair elasticity. The mass was then punctured three times with a fine needle (Figure 2 (d)). Microscopically, the biopsy showed inflammatory exudates and granulation tissue formation (Figure 2(e)). However, TB-PCR was negative. The tissue from the mass puncture showed inflammatory exudates and lymphoid cells (Figure 2(f)).

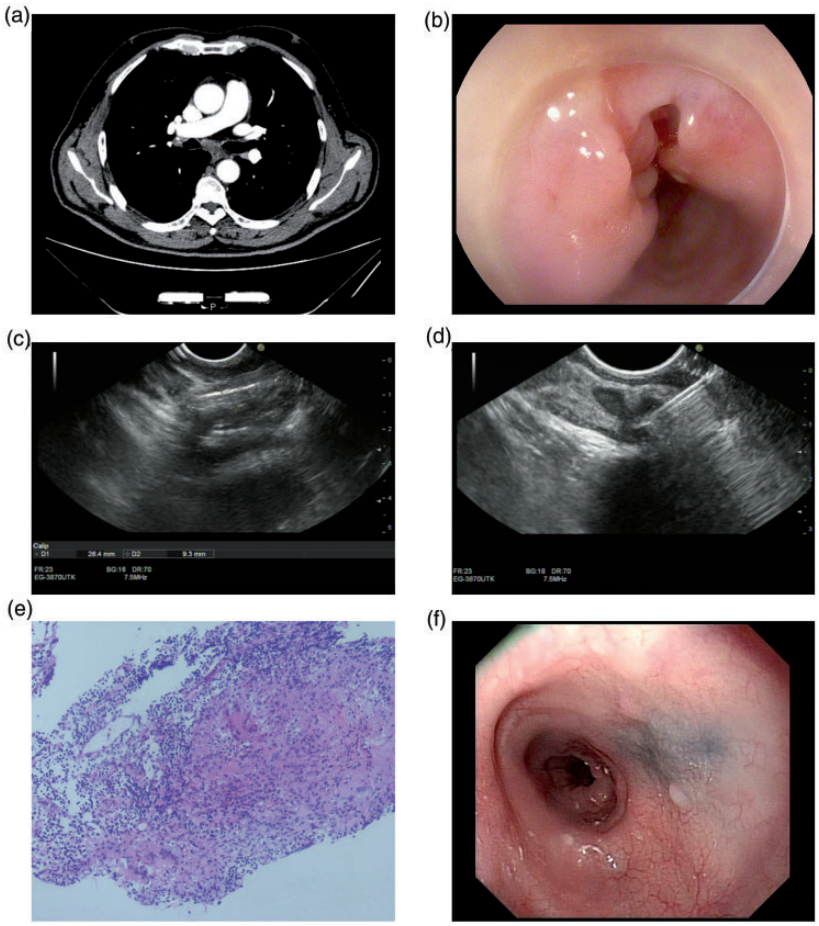

Figure 1. Imaging and endoscopic findings in Case 1. (a) Computed tomography showed enlarged lymph nodes under the carina and an unclear boundary between the lymph nodes and esophagus. (b) Under endoscopy, we found a longitudinal fistula in the esophageal wall. (c) Ultrasound examination showed sinus-like echo near the enlarged lymph node. (d) Ultrasound examination showed multiple lymph nodes under the carina and around the esophagus, and the nodes were punctured with a fine needle. (e) Microscopically, the biopsy showed active chronic inflammation, granulation tissue formation, and local necrosis. (f) Endoscopic re-examination showed that the mucosa of the lesion had healed and that the surface was slightly rough and charcoal black in color.

Based on these clinical manifestations, the patient was clinically diagnosed with esophageal tuberculosis and underwent standard anti-tuberculosis treatment after providing informed consent. For personal reasons, she received treatment and endoscopic review at a local hospital. Her fistula had healed at the 6-month follow-up examination.

The reporting of this study conforms to the CARE guidelines.[6] In this report, we have de-identified all details such that the identity of the patients may not be ascertained in any way.

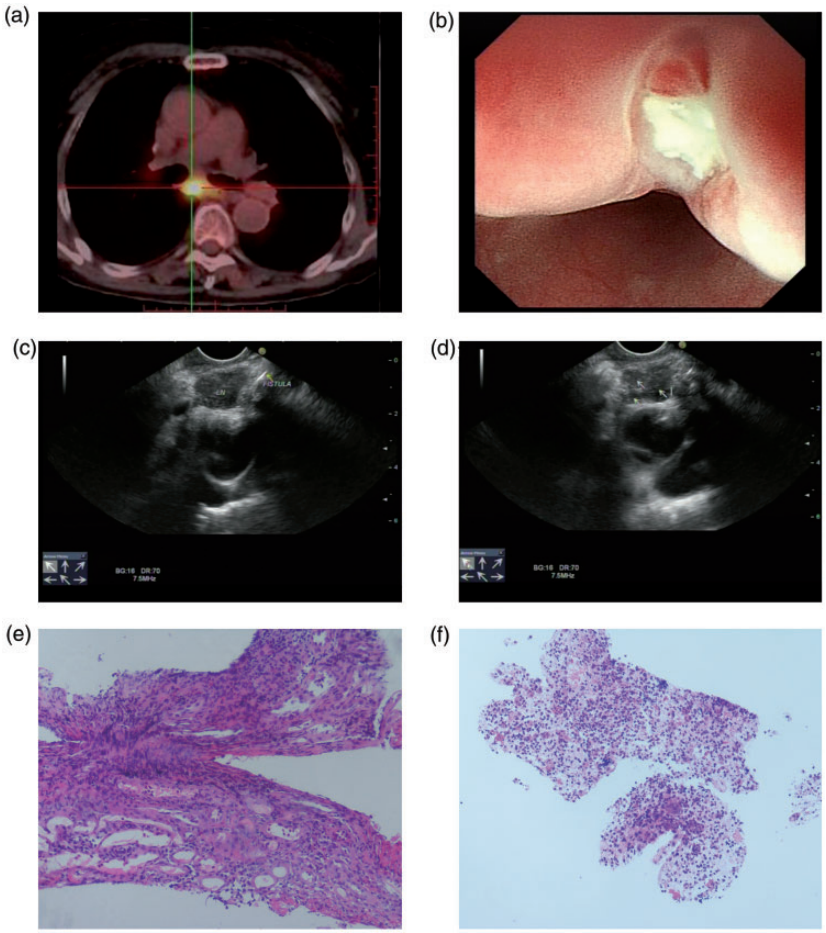

Figure 2. Imaging and endoscopic findings in Case 2. (a) Positron emission tomography–computed tomography showed enlarged lymph nodes under the carina, and the nodes had an unclear boundary with the esophagus and increased fluorodeoxyglucose metabolism. These findings indicated the presence of an esophageal fistula and inflammatory granulomatous lesions. (b) Endoscopic examination revealed a deep ulcer-like lesion in the esophageal wall. (c) Ultrasound examination showed sinus-like echo between the esophageal lesion and the mass. (d) Ultrasound examination showed a hypoechoic mass lesion beside the esophageal ulcer; the internal echo of the lesion was not uniform, and fusion of lesions were seen within it. These lesions were punctured with a fine needle. (e) Microscopically, the biopsy showed inflammatory exudates and granulation tissue formation. (f) The tissue from the mass puncture showed inflammatory exudates and lymphoid cells.

Discussion

Esophageal tuberculosis can be classified as either primary or secondary according to the infection route. Primary esophageal tuberculosis is caused by direct infiltration of sputum containing *M. tuberculosis* and is rare in the clinical setting. This rarity may be associated with multiple protective mechanisms, such as coverage of the esophageal wall by squamous epithelium and the short residence time of food within the esophagus.[7] Secondary esophageal

tuberculosis is the most common type. Previous reports have indicated that esophageal tuberculosis is mainly caused by direct invasion of tuberculous lesions in surrounding tissues or organs (e.g., mediastinal lymph node tuberculosis, thoracic vertebra tuberculosis, and parahilar lymph node tuberculosis).[8–10] Bloodstream infection is rare.[11,12] Esophageal tuberculosis can involve the entire esophagus, but involvement of the middle part is more common.[7] This may be due to the proximity of this region to the carina, where lymph node enlargement is most common. In both of our patients, the lesions were located in the middle part of the esophagus and affected the anterior esophageal wall, which is consistent with the literature. Additionally, chest imaging of both patients showed enlarged mediastinal lymph nodes. These findings suggest that the esophageal lesions in both patients were caused by direct invasion of tuberculosis of mediastinal lymph nodes.

The onset of esophageal tuberculosis is insidious. The main clinical manifestations are chest pain and dysphagia,[1,13] although some patients exhibit tuberculous symptoms such as emaciation[12] and rarely hematemesis.[14,15] One of our patients presented with chest pain and dysphagia, and the other presented with emaciation. This reminds us that we should be alert to this disease when we encounter patients with similar symptoms. In addition, both the PPD test and T-SPOT were positive or strongly positive in our two patients, which is slightly helpful for establishment of the differential diagnoses. It is difficult to differentiate esophageal tuberculosis from a malignant tumor by imaging examination because of the lack of specific imaging signs of esophageal tuberculosis.[16] However, chest computed tomography and other imaging techniques can provide evidence of pulmonary tuberculosis or enlarged mediastinal lymph nodes, thus providing guidance for EUS-FNA.

Through EUS, doctors can not only directly and clearly observe the lesion location, morphology, and type but also carry out real-time ultrasound scanning to obtain information such as the echo situation, edge status of esophageal lesions, and relationship of the lesions with surrounding organs and lymph nodes. In addition to routine biopsy for erosive or ulcerative lesions, EUS can also be used to perform fine-needle aspiration for smooth protruding lesions or enlarged lymph nodes around the esophagus under ultrasound guidance, improving the positive rate of biopsy.[17] The characteristic features of EUS in esophageal tuberculosis are homogeneous or heterogeneous hypoechoic lesions with internal flaking and cord-like hyperechoic lesions, and the outer membrane may be discontinuous. Enlarged lymph nodes can be seen outside the esophageal wall and exhibit fuzzy boundaries, uneven internal echo, and partial fusion; they may be connected to and fused with the esophageal lesions.[12] In our two patients, EUS revealed enlarged lymph nodes outside the esophageal wall. The echogenicity was uneven, fusion was seen, the boundary with the esophageal wall was unclear, and a fistula was found. The discovery of these features is helpful for the diagnosis of esophageal tuberculosis. Endoscopic biopsy and EUS-FNA were performed in both patients. The pathological findings showed granulation tissue formation without evidence of a tumor, which is important for differential diagnosis. In addition, TB-PCR was performed on the pathological tissues of both patients, and the results were negative; notably, this does not support a diagnosis of tuberculosis. However, considering all the clinical manifestations of the patients, we made a preliminary diagnosis of esophageal tuberculosis and administered standard treatment. The therapeutic effect in such cases

can also help us judge whether the diagnosis is correct. After diagnosis, both patients received standardized antituberculosis treatment at a specialized hospital. After treatment, the symptoms gradually decreased in severity and the fistulas healed. The effectiveness of antituberculosis treatment further confirmed the diagnosis of esophageal tuberculosis. These cases illustrate that it is necessary to comprehensively consider the patient's entire condition to achieve the correct diagnosis of esophageal tuberculosis. Notably a limitation in these cases is that we did not send biopsies for tuberculosis culture. If tuberculosis is considered as a differential diagnosis, biopsies should be sent to microbiology for tuberculosis culture.

Conclusion

We have presented two rare cases of esophageal tuberculosis. Both patients presented with an esophagomediastinal fistula, underwent EUS-FNA, received timely antituberculosis treatment, and exhibited clinical improvement.

Authors' contributions

DN conceived of the report and participated in drafting of the manuscript. JL participated in the data acquisition and critically revised the manuscript. WL was responsible for the pathological analysis. PL and SZ carried out the endoscopic maneuvers and participated in the conception and design of the report. All authors read and approved the final manuscript.

Declaration of conflicting interest

The authors declare that there is no conflict of interest.

Ethics statement

Both patients provided informed consent for treatment and publication of this report. All details have been de-identified such that the identity of the patients may not be ascertained in any way. The requirement for review board approval was waived because of the nature of this study (case report).

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