CASE REPORT

Tuberculosis of the esophagus

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SUMMARY. We report a case of a patient with esophageal tuberculosis, a very uncommon form of extrapulnonar tuberculosis. Initially, because of constitutional symptomatology and radiological findings of mediastinal lymph node enlargement, lymphoma was considered. However, the endoscopic findings of ulcerative masses and a sinus tract revealed by esophagram were suspicious of tuberculous origin. Diagnosis was achieved after bacterial examination of smear samples from esophageal ulcers that revealed bacillus tuberculous and histological demonstration of caseating granulomas in cervical lymph nodes. Tuberculous mediastinal lymphadenitis was thought to be source of the spread to esophagus.

The patient was successfully treated with a three antituberculous drugs regimen. In spite of its rarity, even in patients without risk factors, the diagnosis would be considered in the differential diagnosis of uncertain esophageal lesions.

INTRODUCTION

Tuberculosis of the esophagus is a very uncommon condition and usually results from direct extension of caseating mediastinal lymph nodes.¹ Immigrants from undevelopment countries, immunocompromized as well as patients with AIDS are considered at risk. Although 'unique endoscopic appearance' has been described,² the clinical diagnosis, particularly in cases without predisposing factors is in general, difficult. Moreover, the histological diagnosis based on esophageal biopsies is not always definitive³ and often the bacillus is not isolated. Therefore, frequently, the diagnosis is presumed-based in response to anti-tuberculous therapy. Herein we report a patient who was referred to our institution with suspicion of a malignant lymphoma or a malignant submucosal tumor.

CASE REPORT

The patient, a 24-year-old Japanese male student, was referred to our institution with a 3-month history of cough, dysphagia and weight loss of around 3 kg. His past and family history were unremarkable. Physical examination was otherwise normal except for palpable cervical lymph nodes bilaterally. Results of routine hematology and blood biochemistry test were all within normal limits. Chest X-ray revealed bilateral hilar lymphadenopathy without evidence of parenchymal infiltrations, cavities or pleural effusions. Upper endoscopy revealed three apparently submucosal, ulcerated without bleeding masses at 24 cm, 28 cm and 34 cm from the incisors (Fig. 1). Biopsy samples were taken for analysis.

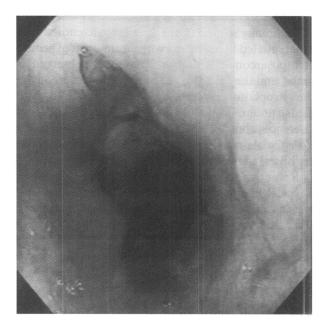


Fig. 1 Endoscopic view of one of the esophageal lesion. Ulceration resembles malignancy although linear configuration would suggest tuberculous origin.

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Chest CT scan showed marked enlargement of upper mediastinal lymph nodes with close contact to the anterolateral aspect of the esophagus (Fig. 2). However, no abnormality was seen in the lung fields. A barium swallow revealed multiple extrinsic compression of the upper-mild esophagus and a sinus tract (Fig. 3). The histological examination of the biopsy specimens were inconclusive demonstrating intense lymphocyte infiltration and marked inflammatory reaction. Bacterial examination of smear samples from esophageal ulcers revealed bacillus tuberculous. Based on these results, biopsy of the cervical lymph nodes was carried out for attempt to confirm diagnosis. The histology confirmed caseating granuloma with epitheloid cell infiltration and Langhans s giant cells. Mantoux test was performed and was found to be positive.

Isoniazid, rifampin and streptomizin were started and 3 months later, symptomatology and cervical lymph nodes had almost disappeared. General conditions of the patient also had improved.

DISCUSSION

Affection of the esophagus by tuberculosis is uncommon and primary as well as secondary forms have been described. The primary form implies absence of tuberculosis in other sites and because stratified squamous mucosa and the rapid transit time protect the esophagus against implantation of tuberculous bacillus, pre-existing esophageal lesions such as reflux esophagitis, inflammation, ulceration, stricture or carcinoma are considered necessary for the implantation of the micro-organism.¹ Cases without pre-existing disorders have been reported^{4,5} although, recently, with the development of acute diagnostic methods, their occurrence has been questioned.⁶ The secondary form is more likely to occur and results of hematoge-

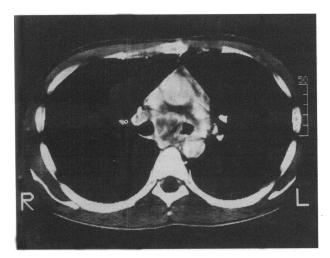


Fig. 2 CT of chest showing enlargement of mediastinal lymph nodes in close contact to the right anterior esophageal wall.

nous or retrograde lymphatic dissemination although caseiting mediastinal lymph nodes are considered the main source of spread. In our case, the patient had evidence of tuberculous mediastinal lymphadenitis as demonstrated by enlargement of paratracheobronchial lymph nodes.

This form of extrapulmonar tuberculosis has been recognized as part of primary disease and has been reported to occur with increasing frequency in adults of developing countries.7 This tendency has been attributed to the reduction of the incidence of tuberculosis and primary infection in children, with subsequent increase of adults susceptible to primary infection.^{5,6} In spite of this and the fact that the esophagus is surrounded by potential source of spread, it is surprising that only around 55 cases of dissemination to esophagus have been reported over the last 75 years,^{8,9} most of them being found in immigrants from undeveloped countries and immunocompromized patients. Due to the rarity of this entity, the diagnosis is not usually suspected and other conditions like lymphoma, sarcoidosis, nonlymphomatous neoplasms including metastatic diseases are primerly considered.

The main complaint of the affected patients consists of dysphagia or odynophagia which are present in around 81% of cases¹⁰ and depend on the presence

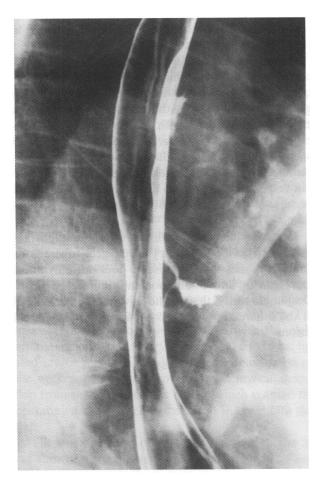


Fig. 3 Esophagram reveals a sinus tract located in the mild thoracic esophagus.

of ulcerative lesions.^{3,11} Systemic symptoms which are non-specific such as fever, weakness and weight loss are reported in most cases.⁶ Exceptional cases, in which the presenting sign has been hematemesis due to esophageal-aortic fistula^{12,13} or erosion of caseating lymph nodes into the esophagus and its blood supply¹⁰ have been also recorded.

The radiological findings are non-specific and consist of ulceration of the mucosa, stricture, pseudotumor masses, fistula and sinus tract formation.⁷ The finding of fistula or esophagomediastinal communication in patients at risk for AIDS are also associated with tuberculous infection.¹⁴ In our case, an esophagram showed a sinus tract formation although patient did not have antecedents associated with AIDS.

The endoscopic appearance ranges from solitary ulcers¹⁵ to nodular, polypoid ulcerative lesions that can resemble malignancy.^{16,17} Ulcerative linear lesions with normal-appearing surrounding esophageal mucosa and fistulous tracts as in our case, have also been described.²

The histological examination of biopsy sample often reveal only changes of esophagitis.¹⁸ Therefore, because findings of casseating granulomas and demonstration of acid-fast bacilli are unusual,^{3,11,19} culture of endoscopic biopsies has been recommended as routine procedure.²⁰ In a significant number of reported cases, the diagnosis of tuberculosis is presumed only after improvement is reached in response to anti-tuberculous therapy.

The recommended regimen consists of isonizid, ethambutol or streptomicin and rifampin with adequate clinical response within 2 to 4 weeks.^{11,19,21} In general, the prognosis of these patients is good.⁵ Even cases complicated with tuberculous tracheoesophageal fistulas have been successfully treated with specific chemotherapy.²² Unfortunately, immunocompromized patients particularly patients with AIDS do not respond adequately to antituberculous treatment because of the high mortality rate within this population.^{14,23}

Our case therefore highlights the more important characteristics of secondary esophageal tuberculosis and exemplifies that this disease does not only affect immigrants and immunocompromized patients. Despite its rarity, this diagnosis would be considered in the differential diagnosis of uncertain esophageal lesions.

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