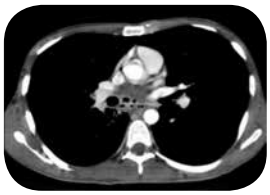

Oesophageal Fistula due to Tuberculosis in HIV Patients: Report of two Cases

Fístula esofágica por tuberculosis en pacientes con VIH: Presentación de dos casos

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Key words (MeSH)

Tuberculosis
Esophageal fistula
HIV

Summary

Extrapulmonary tuberculosis (TB) is frequent in HIV patients; nevertheless, the incidence of esophageal involvement is low and high clinical suspicion is required for a proper diagnostic approach in order to identify Mycobacterium TB as a causative agent of infection. Imaging studies such as conventional chest radiography, esophagogram, multislice computed tomography (MCT), and upper endoscopy provide information based on specific findings that can lead to the diagnosis of TB. They serve as a guide for tissue sampling and confirmatory molecular tests and cultures. This article presents two cases of young male patients diagnosed with HIV/AIDS C3 and co-infected with TB, who developed esophageal and mediastinal lymph node involvement, with secondary perforation and mediastinal fistula.

Palabras clave (DeCS)

Tuberculosis
Fístula esofágica
VIH

Resumen

En los pacientes con VIH es frecuente la tuberculosis (TB) extrapulmonar; sin embargo, la incidencia de afectación esofágica es baja. Se requiere de una alta sospecha clínica para realizar un adecuado enfoque diagnóstico y para identificar al *Mycobacterium TB* como agente causal de infección. Los estudios por imagen, como la radiografía convencional de tórax, el esófagograma, la tomografía computarizada multicorte (TCM) y la endoscopia digestiva superior aportan información basada en hallazgos específicos que pueden orientar hacia el diagnóstico de TB ganglionar y esofágica. Sirven como guía para la toma de muestras de tejidos y la realización de estudios confirmatorios de presencia del bacilo, como las pruebas moleculares y cultivos. Se reseñan 2 casos de pacientes jóvenes, de sexo masculino, con diagnóstico de VIH/sida C3 con coinfección por TB, quienes desarrollaron compromiso ganglionar mediastinal y esofágico, con perforación secundaria y fístula mediastínica.

Introduction

Tuberculosis (TB) represents a global public health problem and has had a secondary resurgence in the AIDS epidemic. In this population group TB is the most frequent opportunistic infection (1,2). The risk of developing it is 50 times greater for an HIV-infected person compared with a healthy one (3).

TB is considered the second cause of death in the world due to communicable diseases, after the human immunodeficiency virus (HIV / AIDS), with an annual mortality of approximately 2 million people. Thirteen percent of patients with TB have coexisting HIV infection (4).

Although lung disease is the most common form of TB, all organs may be affected, especially in the group of patients with compromised immunity (1,2).

In HIV-positive patients, extrapulmonary TB occurs most frequently and its most common clinical manifestation is lymphadenitis, with prevalence close to 35 % (2).

The gastrointestinal tract is the sixth organ in localization frequency of extrapulmonary TB with an incidence of 3-5 % (5). In the latter, the most common location is the ileocecal valve, while the esophagus is one of the least frequent occurrences, with 0.15 to 0.2 % of cases (1,2,6).

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Case 1

28-year-old male patient. He consulted for fever, abdominal pain, dysphagia and diarrhea of several weeks of evolution and hematemesis from the day before admission. During hospitalization he was diagnosed as HIV/AIDS C3 (Elisa and Western Blot Positive, CD4 lymphocyte count of 188).

In multi-slice CT scan (MCT) of thorax with contrast medium, adenomegalias in various ganglionar stages of the mediastinum were found. Through thoracoscopy a lymph node biopsy was performed, which resulted in a positive Ziehl Neelsen (ZN), suggesting the diagnosis of lymph node TB.

Endoscopy of the upper digestive tract was performed in which lesions were found in the esophageal mucosa, suggestive of candida infection. In addition, two ulcers were found in the middle third of the esophagus (Figure 1).

Biopsy and culture of esophageal tissue were negative for TB. During hospitalization, anti-TB management was started. A month after the start of treatment he manifested cough and fever. Dysphagia persisted as well. A MCT was performed with thoracoabdominal contrast medium in which a conglomerate of necrotic mediastinal adenomegalies, with central air content suspect of esophageal fistula and multiple granulomas in both lungs (Figure 2).

Images of the abdomen showed hepatomegaly, splenomegaly with multiple hypodense nodules suggestive of splenic microabscesses; retroperitoneal adenomegaly and a collection in the tail of the pancreas. Subsequently, a contrasted MRI of the abdomen confirmed these findings.

An oesophagogram and fistulogram guided by endoscopy were performed, to evaluate the evolution of esophageal ulcers and to rule out a perforation that explains the tomographic findings of the thorax.

In the endoscopy, a perforated ulcer was found in the third middle of the esophagus. In addition, narrowing of esophageal light due to extrinsic compression secondary to mediastinal adenomegalies was observed (Figure 3).

Contrast medium was injected with a catheter through the ulcer with which a 7 cm fistula was found between the esophagus and the mediastinum in the right subcarinal region (Figure 4).

A new chest MCT scan was performed after the oesophagogram, which showed a fistulous path from the esophagus to the conglomerate of right subcarinal adenomegalias and into the inferior posteromedial pleural space of the right hemithorax (Figure 5).

Three sputum smear microscopies were positive for acid-alcohol resistant bacilli (BAAR). The culture of sputum obtained by bronchoalveolar washing and molecular tests were also positive for isoniazid (H) and rifampicin sensitive TB (R).

The definitive diagnosis was C3 AIDS with disseminated TB disseminated with lymph node, pulmonary and splenic compromise, with perforated scrofula to the esophagus and an oesophagomediastinal and pleural fistula.

Treatment with HAART therapy (highly activity anti retroviral therapy) and a gastrostomy was performed, which was closed a few months later.

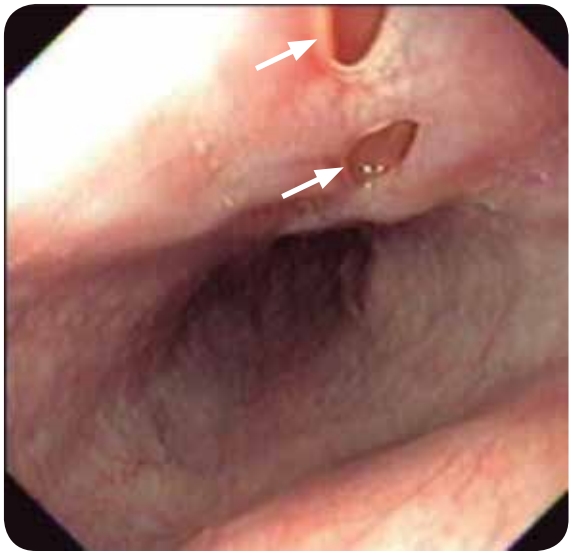

Figure 1. Upper digestive endoscopy. Sample of two esophageal ulcers (arrows). Multiple whitish punctiform lesions adjacent to ulcers, which suggested candida infection.

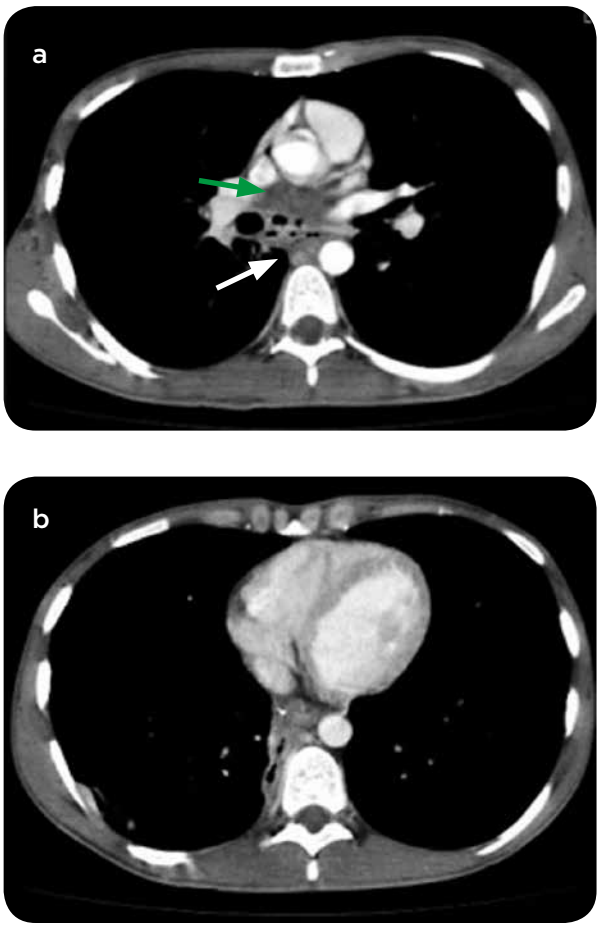

Figure 2. Contrasted thorax MCT scan. a) Air tract by communication of a fistula to the pleural space in the subcarinal location and pleuroacigoesophageal recess (white arrow). Necrotic adenomegaly (green arrow). b) Granulomas in both lungs.

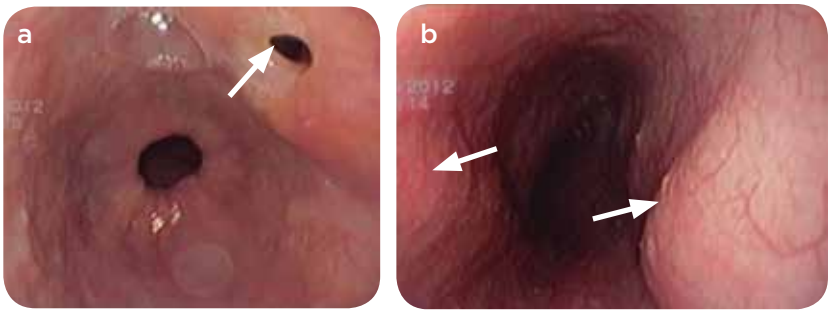

Figure 3. Upper digestive endoscopy. a) Ulcer and perforation in the middle third of the esophagus through which contrast medium is passed through a catheter (arrow). b) Mediastinal adenomegalias that produce compression of the light of the middle third of the esophagus (arrows).

Case 2

27-year-old male patient. 6-month history of dysphagia, heartburn, wet cough, adynamia, nocturnal diaphoresis, loss of 15 kg of weight and multiple cervical and axillary adenomegalias.

Upper digestive endoscopy showed antral gastropathy, active bulbar duodenitis and biliary gastric duodenum reflux. The gastric biopsy was positive for Helicobacter Pylori. He received antibiotic treatment. Five Months later he consulted with fever, headache, dysphagia and persistence of cervical adenomegalias.

A diagnosis of HIV-AIDS C3 was made with positive Elisa and Western Blot reports. CD4 lymphocyte count of 142 cells/mm^3 and viral loading of 22,335 copies.

A lumbar puncture was performed and analysis of the cerebrospinal fluid (CSF) reported glucose of 54 mg/dL, proteins of 42 mg/dL, leukocytes 0, no bacteria; Adenosine diamine (ADA) tests and polymerase chain reaction (PCR) for TB, negative; latex and chinese ink for Cryptococcus, negatives.

A cervical ganglion biopsy was performed. The pathology reported the presence of caseous center, positive PCR for TB, sensitive to R, with negative ZN and KOH.

The chest X-ray showed paratracheal mediastinal enlargement, increased volume of right pulmonary hilum and of the pleuroacid esophagus recess, with lower paraillary reticulonodular opacities (Figure 6).

Contrast CT scan of the chest showed multiple right inferior mediastinal adenomegalies with necrotic center suggestive of TB compromise and a fistulous pathway was defined with air content between the esophagus and the right subcarinal adenomegaly. As well as a pattern ofa budding tree with multiple centrilobulillary nodules in the inferior right lobe (Figure 7).

MCT scan with contrast medium of the abdomen showed multiple mesenteric adenomegalies with necrotic center suggestive of compromise by TB (Figure 8).

MCT scan with neck contrast medium showed adenomegaly in several cervical ganglionic chains, also with a necrotic center (Figure 9).

A new endoscopy of the upper digestive tract was performed where an extensive ulcer with perforation in the middle third of the esophagus was found. It was not possible to determine its depth. A great amount of pus was vacuumed that left a crater compatible with oesophagomediastinal fistula. Histopathology reported a positive PCR for TB.

The definitive diagnosis was C3 AIDS with disseminated TB, with pulmonary, cervical-mediastinal and mesenteric ganglion compromise, perforated scrofula to the esophagus and esophageal-mediastinal fistula.

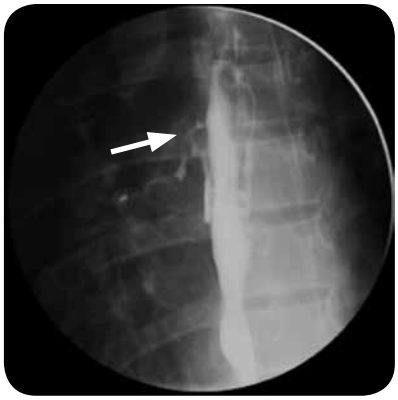

Figure 4. Image of oesophagogram-fistulogram. Fistula of 7 cm of length between the middle third of the esophagus and the mediastinum in the right subcarinal region (arrow)

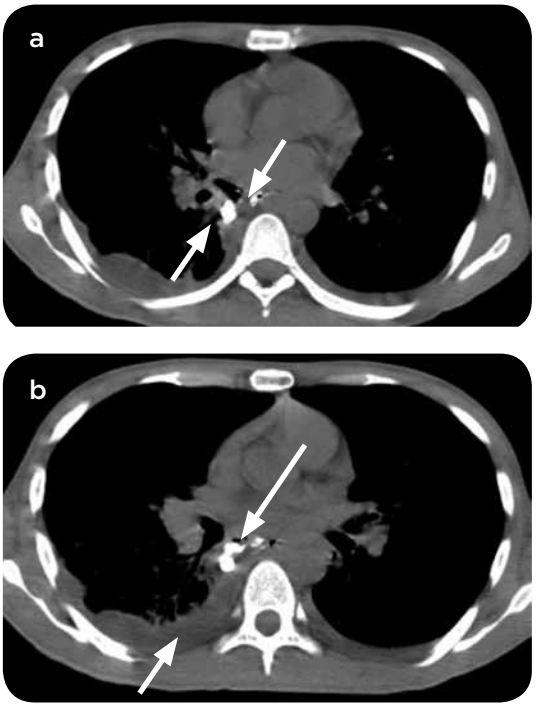

Figure 5. Chest MCT. a) Fistulous path from the esophagus to the right subcarinal adenomegalias conglomerate and into the lower posteromedial pleural space of the right hemithorax, with contrast medium injected into the fistulogram (arrows). b) Medium of contrast by the fistulous path (long arrow) and pleural effusion right (short arrow)

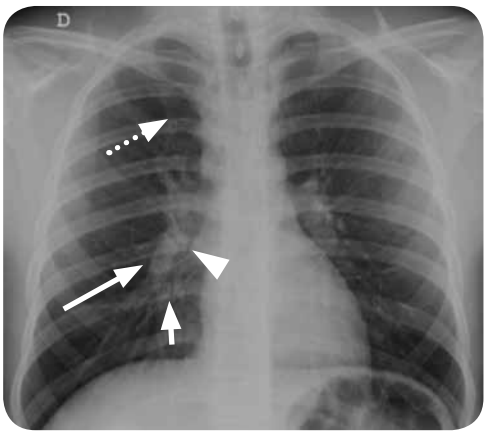

Figure 6. PA chest X-ray. Right paratracheal mediastinal enlargement (dotted arrow). Increased volume of right pulmonary hilum (long arrow) and pleuroacisoesophageal recess (arrow head). Mixed lower right paraillary opacities (short arrow).

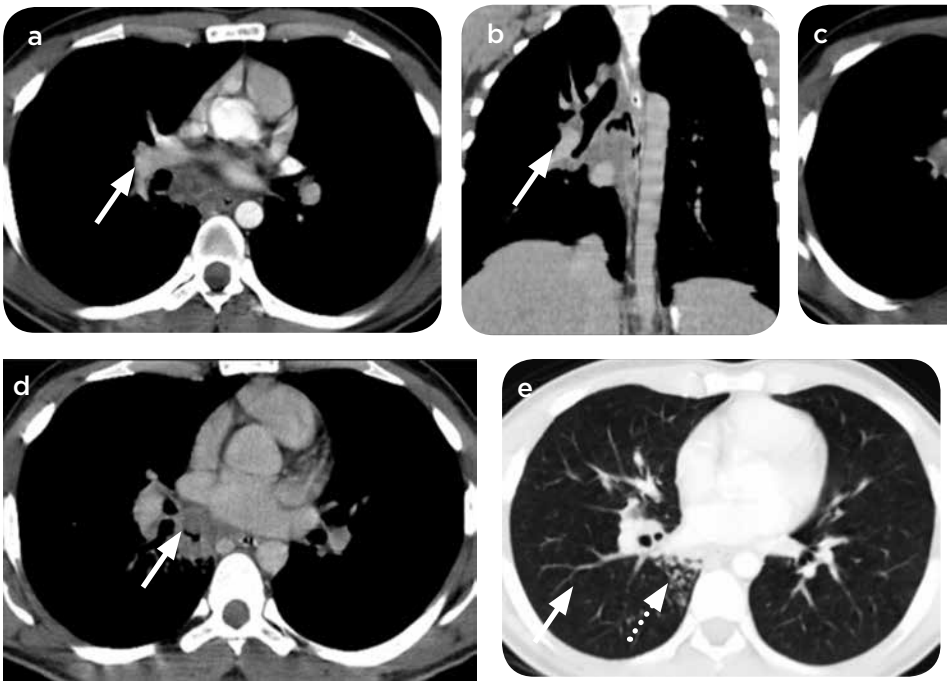

Figure 7. Contrasted MCT chest scan. a) MCT axial section. Subcarinal adenomegalies with necrotic center and peripheral enhancement, suggestive findings of compromise for TB (arrow). b) MCT coronal slice. Necrotic adenomegalies of right paratracheal predominance, subcarinal and in the pleuroacigoesophageal recess (arrow). c) Fistulous pathway with air content between the esophagus and right subcarinal adenomegalies (arrow head). d) Conglomerate of adenomegalies in pleuroacigoesophageal recess. With central air presence of a fistula (arrow). e) MCT of axial thorax. Centrilobulillary nodules in the lower lobe of the right lung (head of arrow). Budding tree pattern (arrow).

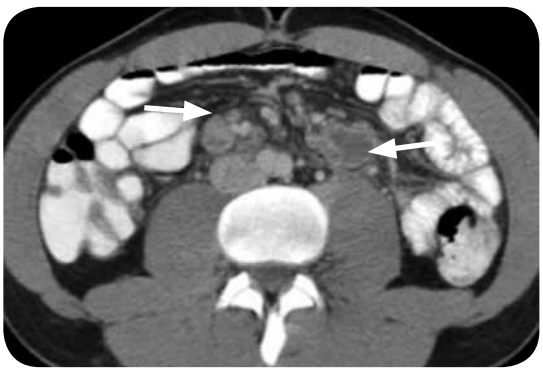

Figure 8. MCT with contrast medium of abdomen. Mesenteric adenomegalias with necrotic center (arrows).

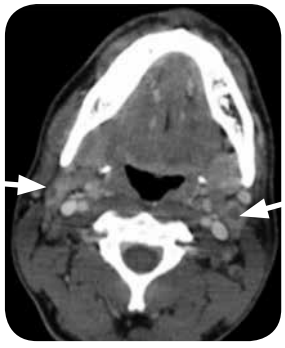

Figure 9. Cervical MCT with contrast medium. Necrotic adenomegaly with center suggestive of TB compromise (arrows).

Discussion

Esophageal TB is usually due to a local extension of mediastinal adenomegalies that produce extrinsic compression of its wall. This phenomenon occurs mainly in the third middle of the esophagus, at the level of carina (2,6-8).

In MCT, the ganglionic involvement due to TB is characterized by low central attenuation indicating necrosis and peripheral enhancement in ring (1,7,8).

Primary esophageal TB due to colonization of the bacillus in the mucosa when swallowing sputum is rare. There are protective mechanisms, such as saliva, squamous epithelium, peristalsis, and an oesophagic sphincter that prevent reflux (8,9). The hematogenous and lymphatic retrograde spread has also been described, but the latter are very rare (5,10).

Concomitant pulmonary disease occurs in 25 % of patient's cases of gastrointestinal TB. The main clinical manifestation of esophageal TB is dysphagia (5,8,10-12). It occurs in 90 % of the cases (10) due to the presence of intrinsic ulcers, tracheoesophageal fistula or extrinsic compression by mediastinal or cervical nodules (5). Other associated symptoms are odynophagia, retrosternal pain, cough during swallowing in fistulizing presentations, hematemesis, fever and weight loss (5,10,12).

The clinical picture is similar to that of other esophageal pathologies, which delays the timely diagnosis and treatment. In some cases it can also be confused with esophageal carcinoma and this must considered as part of the differential diagnosis (5,10).

The diagnosis of esophageal TB requires a high index of suspicion in patients with a history of lymph node or pulmonary TB which present esophageal ulcers. The most frequent endoscopic finding of esophageal TB is the solitary ulcer with an excavated base and raised edges (5).

In these cases it is recommended to perform an oesophagoscopy or endoscopy with additional sampling routine for confirmation of Mycobacterium Tuberculosis or of caseifiyng granulomas. Resistant acid-alcohol staining is positive in less than 25 % of the cases (5,9); for this reason, culture for mycobacteria, which has a sensitivity > 80 % and a specificity of 98 % must be performed.

Several clinical trials have shown that PCR for Mycobacterium TB is currently a test of great value and allows a quick diagnosis. It has a sensitivity and specificity similar to conventional culture techniques (13).

A thorax MCT should always be performed with contrast medium to differentiate primary esophageal TB from secondary. The latter is defined by adenomegalies that suggest tuberculous lymphadenitis. The MCT also ruled out lung TB (5) and complications such as perforation, pneumomediastinum and Pott's disease (13).

Oesophagomediastinum fistulas are a rare complication of esophageal TB (9,11); they are produced by erosion of the wall of the esophagus by contiguity of a mediastinal adenomegaly during active tuberculous adenitis or secondary to pneumonia due to TB. Also, they can present by calcification of an adenomegaly that erodes and produces a secondary broncolite (9).

Esophagitis in retroviral disease can also cause ulceration, perforation and formation of fistulas (14).

The finding by MCT of oesophagomediastinum fistulas is of a linear image with air density, located in the periphery of a mediastinal lymph node, mainly on the right side (80 %) and toward the intermediate bronchus (11).

The 'sign of Ono' is pathognomonic of oesophagomediastinal fistula, includes paroxysmal coughing by ingesting liquids and crepitation over the sixth right posterior intercostal space (9). After the scarring process of the fistula occurs, traction diverticuli may appear in sites where fistulas were previously identified (9,15).

With respect to treatment, case series and other studies have demonstrated that medical treatment with anti-TB drugs has been favorable in the resolution of esophagomediastinal fistulas (9,10).

In the case of immunocompetent patients, standard tuberculostatic therapy for a period of 6 to 9 months is applied (5). Conservative treatment with prostheses and anti-TB therapy, in general, are successful (9,10).

In the two cases presented, the patients were diagnosed with TB confirmed by PCR or culture, associated with HIV/AIDS. Clinically, both patients presented dysphagia as a symptom associated which led to hospitalization.

In the imaging studies we documented the finding of mediastinal adenomegalias with compromised contiguity of the esophagus and perforation with secondary mediastinal fistula.

Endoscopy made it possible to diagnose esophageal ulcers and the fistulous trajectory was documented by contrasted MCT scan of the thorax. Both received anti-TB medical treatment with resolution of the infection and cure of the oesophagomediastinal fistula.

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