

Case Report

Oesophageal and Oral Ulcer Tuberculous Associated with Human T-cell Lymphotropic Virus Type I (HTLV-I) and SARS-COV-2 Infection

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DOI: <https://doi.org/10.24321/0019.5138.202312>

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How to cite this article:

Montalvo R, Vilcapoma P, Pomazongo M, Mathey C, Montalvo M. Oesophageal and Oral Ulcer Tuberculous Associated with Human T-cell Lymphotropic Virus Type 1 (HTLV-1) and SARS-COV-2 Infection. *J Commun Dis.* 2023;55(1):78-81.

Date of Submission: 2023-03-10

Date of Acceptance: 2023-04-21

A B S T R A C T

Oesophageal involvement due to tuberculosis (TB) and coinfection with HTLV-1 is rare and can be complicated by the presence of other infections that affect the mucosa of the upper respiratory tract, such as COVID-19. We present the case of a 27-year-old male patient, with 3 months of illness, weight loss, dysphagia, and ulcers in the oral cavity. Tomography showed oesophageal perforation and after oesophageal ulcer biopsy, TB was diagnosed, in addition to infection by HTLV-1 and severe acute respiratory syndrome coronavirus 2 (SARS-COV-2). The patient responded satisfactorily to antituberculous treatment and corticosteroids. Considering the association between rare extrapulmonary tuberculosis and other immunosuppressive pathologies, it is crucial to identify these pathologies in such patients.

Keywords: Tuberculosis, Oesophageal Ulcer, SARS-COV-2, COVID-19, HTLV-1

Introduction

Tuberculosis (TB) is a chronic infection that in most cases produces a latent non-contagious disease because *Mycobacterium tuberculosis* is suppressed by the immune response. Only approximately 5% of infected individuals may develop an active disease in the following years.¹ Extrapulmonary involvement is rare, accounting for about 15% of all TB cases, of which oral and oesophageal involvement usually represent 0.05% to 5%.^{2,3}

The reactivation of tuberculosis is usually due to

immunosuppression processes triggered by debilitating diseases caused by viruses such as human immunodeficiency virus⁴ or HTLV, which generally cause multiple simultaneous opportunistic pathologies due to a decrease in the cellular immune response mainly of the Th2 type CD4 lymphocytes⁵ and other viral infectious processes such as SARS-COV-2, which can precipitate severe manifestations of the disease.⁶

We present here a case of a patient with HTLV-1 and SARS-COV-2 infection, who presented with oral ulcers and oesophageal perforation due to tuberculosis.

Case

A 27-year-old male patient, from the Andes of Peru, with no history of travel to tropical regions presented with a history of weight loss in the last 3 months from 60 kg to 40 kg, associated with dysphagia to solid foods, in addition to evidence of ulcerative lesion on the hard palate and tongue in the last two months, without apparent cause. The lesion was painful on mobilisation and contact with food. In the last week, he presented with fever, general malaise, and a persistent dry cough. He went to the hospital emergency room due to retro sternal pain experienced in the last two days.

Physical examination revealed an ulcer on the edge of the tongue and another on the palate with regular edges, firm consistency, and pain on mobilisation; there was no evidence of lymph node involvement (Figure 1). Pulmonary examination revealed crackles at the base of the right hemithorax and excavated abdomen with a positive urachus sign. The complete blood count (CBC) showed lymphopenia and eosinopenia. The immunochromatographic IgM and IgG tests for COVID-19 were positive. Tests for human immunodeficiency virus, hepatitis C virus, hepatitis B virus, herpes virus 1-2-5, syphilis, fungal cultures, sputum and stool smears, urine lipoarabinomannan antigen, and parasitological examination revealed negative results. Finally, the ELISA test for HTLV-1 was positive.

The neck tomography revealed perforation of the oesophagus

(Figure 1a). Based on the examination, the patient was diagnosed with a perforated oral and oesophageal ulcer, along with COVID-19 and HTLV-1 infection.

The initial treatment was intravenous dexamethasone 6 mg once a day for 10 days, acetylcysteine 600 mg orally 3 times a day for 7 days, ivermectin 40 drops orally once a day for 2 days, and azithromycin 500 mg orally once a day for 3 days; all this was part of the treatment protocol for COVID-19. No evidence of improvement of the oral ulcer was found.

The patient underwent upper gastrointestinal endoscopy, which revealed an ulcer of approximately 1 cm in diameter with regular edges and a whitish erythematous base, an incisional biopsy of the oesophageal ulcer was performed under local anaesthesia (2% lidocaine with 1:100,000 epinephrine). In the pathology study, multiple acid-fast bacilli were identified by Ziehl Neelsen staining (Figure 1b). Investigations were done to rule out the presence of oesophageal and oral cancer in the samples obtained by biopsy, which revealed a negative result.

Established according to the histological findings, the oral and oesophageal ulcer was finally diagnosed as oesophageal TB, along with coinfection by SARS-COV-2 and HTLV-1. The treatment was started with isoniazid, rifampicin, pyrazinamide, and ethambutol with dosage according to weight. The patient showed improvement with greater oral tolerance and was discharged due to favourable progression.

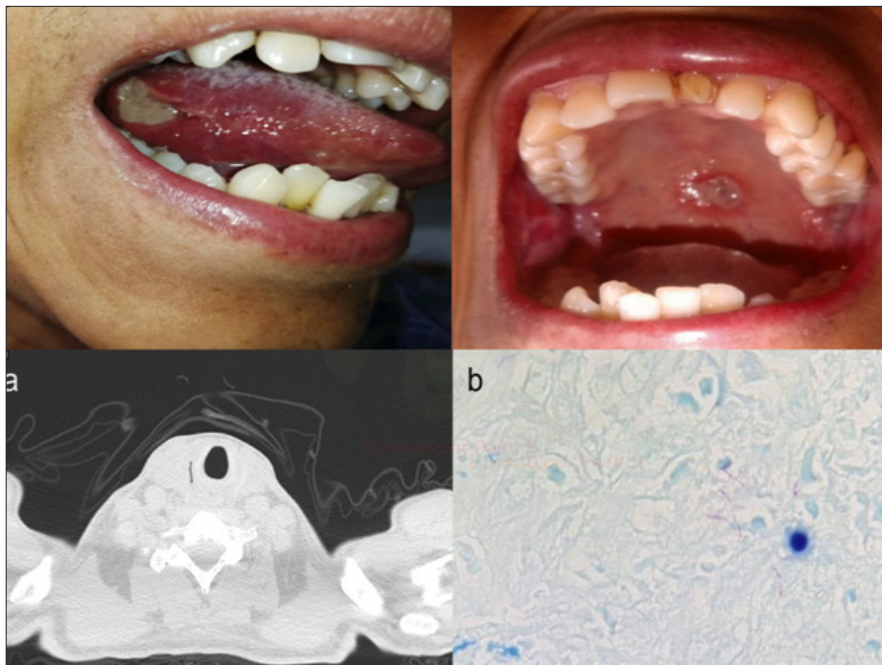


Figure 1. Lingual and Palate Ulcer with Defined Edges and Whitish Base. (a) Cervical Tomography showing the presence of Linear Hypodensity in the Region of the Right Oesophagus, compatible with Oesophageal Perforation. (b) Histopathology showing Acid-alcohol-fast Bacillus

Discussion

Tuberculosis is a disease of worldwide distribution with high mortality,⁷ mainly affecting people who suffer from some type of immunosuppression such as malnutrition, HIV infection, cancer, or as in this case, HTLV-1 infection. The clinical presentations can be very diverse, making the diagnosis more difficult,⁸ mainly in the current context of the COVID-19 pandemic, where many hospitals have neglected other infectious pathologies such as tuberculosis and other respiratory diseases.⁹

Tuberculosis can affect any organ and system, but oesophageal involvement is usually a rare presentation of the disease. Mycobacterium can enter the oesophageal mucosa when ingested from respiratory tract secretions, hematogenous dissemination or by direct contact of contiguous focus.¹⁰ These lesions usually appear as reactivation of latent foci. These oral or oesophageal cavity lesions produce granulomatous inflammation that later ulcerates, which may be complicated by bleeding or perforation, as in the presented case. Symptoms of oral and oesophageal tuberculosis are generally non-specific, manifesting as mild ulcers that can mimic other malignant or fungal conditions¹¹; some general symptoms such as night sweats, decreased appetite, and weight loss are common features. Radiological examination such as oesophageal tomography helps to show the invasion of other tissues and confirm complications such as perforation as evidenced in the patient.

Endoscopic methods are important for diagnosis and for obtaining the sample. These biopsied tissues must be sent for microbiological study for culture of fungi and mycobacteria,¹² however in the presented case, the sample obtained was only sent for pathological anatomy study, and was stained using the modified Ziehl-Neelsen technique. Alcohol-resistant bacilli, compatible with *Mycobacterium tuberculosis*, were evidenced. It was not possible to identify the mycobacterium species, but based on good clinical response and resolution of symptoms with anti-tuberculosis treatment, we arrived at the diagnosis of an oesophageal ulcer associated with tuberculosis.¹³ The Ziehl-Neelsen technique has a sensitivity of 75% for the diagnosis of extrapulmonary tuberculosis.¹⁴

The perforation of the oesophageal ulcer could be triggered by bouts of coughing and local inflammation caused by the SARS-COV-2 virus that affected the patient and led to the patient being admitted to the hospital emergency room. Currently, many infectious pathologies such as tuberculosis can be exacerbated or complicated by the presence of SARS-COV-2, due to the inflammation of the respiratory tract and frequent coughing spells which may also unmask latent pathologies such as HTLV or HIV infection. Hence

it is important to look for coinfections in such patients considering the evidence of rare diseases.¹⁵

Conclusion

Oral and oesophageal tuberculosis is a disease with common characteristics such as chronic ulcer, which generally occurs in patients with some type of immunosuppression, like HTLV-1 infection, as seen in the presented case. This case showed the presence of BAAR bacilli in the histological study obtained by biopsy of the oesophageal ulcer. In the current pandemic, it is imperative to rule out SARS-COV-2 co-infection, which could cause oesophageal perforation due to local inflammation and coughing spells. Thus it is important to identify such associations.

Conflict of Interest: None

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