

Secondary Lingual Tuberculosis Resembling Lingual Cancer with Lung Metastasis: Case Report

Akciğer Metastazı Olan Dil Kanserini Taklit Eden Sekonder Lingual Tüberküloz: Olgu Sunumu

*Kürşat CEYLAN, MD, **Hülya BAYIZ, MD, *Ahmet YAVANOGLU, MD, *Zeynep KIZILKAYA, MD,
*Erdal SAMİM, MD, ***Hüseyin ÜSTÜN, MD

* Ministry of Health Ankara Training and Research Hospital, E.N.T. Department, Ankara

** Ministry of Health Ankara Atatürk Thoracic Diseases Training and Research Hospital, Pulmonology Department, Ankara

*** Ministry of Health Ankara Training and Research Hospital, Pathology Department, Ankara

ABSTRACT

Tuberculosis of the tongue is a rare condition that is usually associated with pulmonary tuberculosis. We present a case of secondary lingual tuberculosis in a 51-year-old man, heavy cigarette smoker and alcoholic who had disseminated lesions in lungs and had ulceration and infiltration of the tongue. Treatment with antibiotics was ineffective. He was admitted to the otolaryngology department because of suspicion of the tongue cancer. Although lingual dorsum is not a frequent localization for the lingual cancer, painful, necrotic, granular, friable and ulcerative nature of the lesion made lingual cancer to be taken into account in the differential diagnosis. The clinical manifestations, diagnosis and the response to the antituberculosis treatment are considered with the literature review.

Keywords

Tongue, tuberculosis, lingual cancer, lung metastasis

ÖZET

Dil tüberkülozu genellikle pulmoner tüberküloz ile birlikte görülen nadir bir klinik durumdur. Bu yazıda ağır sigara içicisi, alkolik ve dilde ülserasyon ve infiltrasyon ile birlikte yaygın pulmoner lezyonları olan 51 yaşında bir erkek hasta rapor edilmiştir. Antibiyotikler ile tedavi yanıtı kalmıştır. Hasta dil kanserinden şüphe edilerek kulak burun boğaz kliniğine refere edilmiştir. Dil dorsumu lingual kanser için mutad bir yerleşim yeri olmamasına karşın lezyonun ağrılı, nekrotik gamüler, frajil ve ülseratif karakterleri dil kanseri şüphesini doğurmuştur. Klinik bulgular, ayırıcı tanı ve antitüberküloz tedaviye cevap hususları literatür bilgileri ışığında tartışılmıştır.

Anahtar Sözcükler

Dil, tüberküloz, lingual kanser, pulmoner metastaz

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Correspondence

Dr. Kürşat CEYLAN

4.Sok. Manolya Apt. 22/27 Sogutozu, ANKARA, TURKEY

Tel: 0532 335 47 97

E-posta: ceylankursat@ttnet.net.tr

INTRODUCTION

Tuberculosis (TBC) still represents a major public health problem worldwide. The incidence of TBC has recently increased as a result of its association with AIDS. About 95% of the individuals exposed to *Mycobacterium tuberculosis* remain clinically asymptomatic, while 5% develop disease. This primary form of the disease is most often localized to the lungs. In most patients the infection does not spread and, as host immunity develops, the caseous foci in the lungs and hiliary nodes undergo healing by fibrosis and eventually calcification. In a minority of patients, progressive pulmonary disease spreads to other organ systems through self inoculation via infected sputum, blood or lymphatic system, establishing the secondary form of TBC.^{1,2}

Oral TBC lesions are infrequent; it is estimated that only 0.05-5% of total TBC cases may present with oral manifestations.³ The most common site of oral TBC is the tongue, however, other oral sites may also be affected.^{1,3} Involvement of the oral cavity by TBC can represent primary or, more often, secondary disease.⁴ Generally occurring in younger patients, the lesions of primary oral TBC are often associated with caseation of the regional lymph nodes and remain painless in the majority of the cases. On the other hand, secondary lesions are more commonly seen in older individuals.⁴ Oral lesions of tuberculosis are nonspecific in their clinical presentation and are often not considered in the differential diagnosis, especially when oral lesions are present before systemic symptoms become apparent.^{5,6}

In this paper we present a case of secondary lingual tuberculosis with miliary tuberculosis, emphasizing its resemblance to cancer of the tongue with lung metastasis.

CASE REPORT

A 43-year-old man referred to our clinic in January 2006 with a two-month history of progressive painful swelling of the posterior one-third of the tongue. During that period he had lost 15 kg of his body weight due to severe odinophagia and there was a history cough, night sweating and pyrexia. He was a heavy cigarette smoker and an alcoholic. He had been treated for glossitis and tongue ulceration with systemic antibiotics and local medication with no improvement. He was referred to our clinic because of suspi-

cion of the tongue cancer. Malnutrition and dehydration was observed and palpation of the neck revealed bilateral enlarged, nontender submandibular and upper jugular lymph nodes.

Oral examination revealed poor oral hygiene, the absence of several teeth, and extensive carious lesions involving many of the remaining teeth. A painful, necrotic, granular, friable ulcer with 2 cm diameter was present on the posterior one-third of the tongue on the midline. The ulcer had an irregular periphery and elevated borders but minimal induration (Figure 1).

The initial clinical impression was that the lesion was squamous cell carcinoma. Incisional biopsy of the ulcer revealed granulomatous inflammation containing Langhans-type giant cells (Figure 2). Periodic acid-Schiff and Gracot-Gomori stains were negative for fungi. Acid-fast bacilli were found in the sections when tissues were stained with Ziehl-Neelsen method.

The patient was hospitalized for further diagnostic evaluation. Chest tomography revealed fluffy and stringy densities in the upper and middle portions of the both lungs (Figure 3). Sputum cultures showed acid-fast bacilli only at the fourth collection and were negative for *Paracoccidioides brasiliensis*. The results of serologic tests for *Paracoccidioidomycosis* and human im-



Figure 1. A necrotic, granular, friable ulcer with 2 cm diameter was present on the posterior one-third of the tongue on the midline. The ulcer had an irregular periphery and elevated borders but minimal induration.

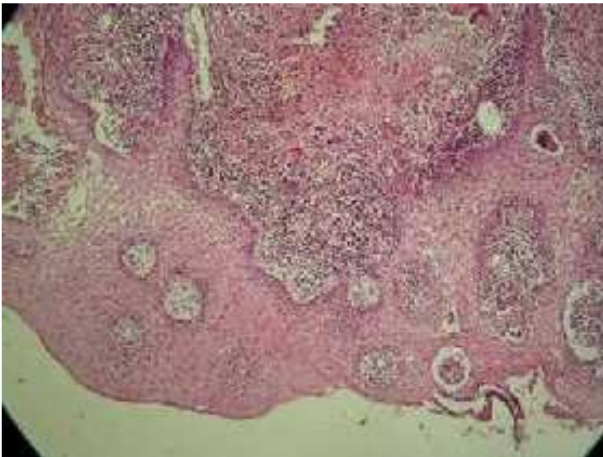


Figure 2. Langhans giant cells and granulomatous structures surrounded with epithelioid histiocytes under hyperplastic squamous epithelium (H&E, 50x).



Figure 3. Chest tomography revealed fluffy and stringy densities in the left and right lungs.

munodeficiency virus were also negative. As a result laboratory data confirmed the histopathological diagnosis.

An antituberculosis therapeutic regimen consisting of rifampicin, isoniazid and pyrazinamide was instituted. The patient showed marked improvement after six weeks, regression of the lung lesions was achieved and the tongue ulcer healed within ten weeks. His last follow-up examination in July 2006 showed normal appearing tongue mucosa with some residual scarring. Antituberculous therapy for one year was advised.

DISCUSSION

Oral TBC lesions may be either primary or secondary, although there are atypical cases reported in the literature. The tongue is the most common site of oral involvement where the tuberculous lesions usually present as chronic irregular ulceration, while an exophytic granular or nodular mass constitutes a less common clinical presentation.^{1,3,7,8} Combinations of these patterns are possible, as exemplified by the case presented here, which featured both deep irregular ulcerations and areas of granularity. The differential diagnosis of a deep ulcerated and/or granular lesion in a lingual location should include reactive and traumatic lesions, malignant tumors, especially squamous cell carcinoma, deep fungal infections, including paracoccidioidomycosis and histoplasmosis and oral manifestations of systemic diseases, such as sarcoidosis and Wegener's granulomatosis.⁷ In the case presented here lingual cancer was suspected before referral to our clinic. Although lingual dorsum is not a frequent localization for the lingual cancer, painful, necrotic, granular, friable and ulcerative nature of the lesion made lingual cancer to be taken into account in the differential diagnosis. The granulomatous reaction observed on histopathological examination was in accord with a potential fungal infection; however, no fungal organisms were identified in the biopsy material, thus ruling out this possibility along with the serologic tests.

Oral TBC lesions are characterized by severe, unremitting and progressive pain that interferes seriously with proper nutrition and rest. However, with the decreased incidence of TBC, the unusual forms oral TBC became unlikely to be diagnosed. Although the pain is greatly reduced within few days after the introduction of chemotherapy, the ulcerations and fissures usually take a few weeks to resolve.^{9,10} The exact diagnosis in our case was made with positive sputum culture confronting the histopathological examination of incisional biopsy of the tongue lesion.

The clinicians should be alert of the possibility of TBC in the differential diagnosis of atypical lesions of the oral cavity, including those appearing as ulcerated and/or granular lesions in a lingual location.

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