Tuberculosis of the Tongue with Coexistent Squamous Cell Carcinoma: An Interesting Case Presentation

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ABSTRACT
Tuberculosis of the tongue is a rarity even in tubercular endemic areas. The disease has a varied clinical picture, most of which may mimic malignant lingual neoplasms clinically. We present a rare case of a 30 year old male patient with complaints of an ulcerated lingual lesion, clinically suspected to be malignant which after histopathological examination was diagnosed as lingual tuberculosis with squamous cell carcinoma.

Keywords: Carcinoma; Glossitis; Tongue; Tuberculosis

INTRODUCTION
Tuberculosis is an infectious disease, caused by an acid fast bacillus, Mycobacterium tuberculosis which has aero-nasal transmission. The disease affects approximately 8 million people worldwide with a relatively high incidence in the Indian subcontinent, South East Asia and Africa [1]. Tuberculosis of the oral cavity is a rare entity and rarely features in the differential diagnosis of oral lesions. The outbreak of drug-resistant TB added with the emergence of acquired immune deficiency syndrome has led to a rise in oral manifestations of tuberculosis [2].

Tuberculosis is a major cause of ill health and death worldwide. Tuberculosis of the tongue, whether primary or secondary, is quite a rare phenomenon [3-5]. The clinical diagnosis is fraught with difficulty as it is rare to suspect that an ulcer, fissure or nodular mass in the tongue is tuberculous in origin. Mostly, the diagnosis is made on fine needle aspiration cytology or histopathologic biopsy from the lesion. We report a rare case of lingual tuberculosis with coexistent squamous cell carcinoma in a 30-year-old male who presented with ulcer in the right antero-lateral surface of the tongue, which illustrates the difficulty in making the diagnosis of extrapulmonary TB with carcinoma and emphasizes the need of a high index of suspicion in such lesions.

CASE SUMMARY
A 30-year-old male presented to the oral surgery unit with complaints of a non-healing ulcer on the right antero-lateral surface to the tongue since the last 4 months despite of administration of antibiotics and topical medicated mouthwashes. He also noticed increasing hoarseness and decrease in appetite with some weight loss for the last 2 months. There was no history of fever, constant cough with expectoration or haemoptysis. The patient was from a low socioeconomic status and a chronic tobacco smoker with gutka chewing for the last 10 years.

On examination, the patient was thin built, appeared pale and unwell. He had an ulcerated lesion of the right antero-lateral surface of the tongue, 3 cm x 1 cm in size with a shallow ulcerated base and rolled margins with poor oral hygiene. There was no evidence of any lymphadenopathy. A strong clinical suspicion of malignancy was elicited and an urgent incisional biopsy was performed under local anaesthesia. Microscopic examination revealed an inflamed ulcerated lesion with several epithelioid cell granulomas with langhan's giant cells (Figure 1) and irregular sheets of highly atypical squamoid cells with overlying acanthotic stratified squamous epithelium (Figures 2 & 3). Subsequent stains for fungi (PAS) and bacteria (Gram stain) were negative. However several acid-fast bacilli were identified with Ziehl-Neelsen stain (Figure 4) leading to a diagnosis of ulcerated lingual tuberculosis with squamous cell carcinoma.

Blood tests were within normal limits except for a raised white cell count with lymphocytosis, a raised erythrocyte sedimentation rate (96 mm in 1st hour) and a raised C-reactive protein (77.6 mg/L). Chest x-ray showed a minor degree of consolidation and cavitation in the right upper lobe, consistent with tuberculosis. He was prescribed 4 drugs anti-tubercular treatment of Rifampicin, Isoniazid, Ethambutol and Pyrazinamide for 9 months with Co-60 radiation therapy x 25 fractions and adjuvant cisplatin chemotherapy (50 mg/m2 x 6 cycles). After 6 months of follow up period, the condition of the patient has improved with complete resolution of the lingual lesion.

DISCUSSION
Tuberculous lesions of the oral cavity may be either primary or secondary to pulmonary tuberculosis, with secondary lesions being more common [6]. The rarity of oral tuberculosis is due to the continuous cleansing of the oral mucosa by saliva and to the presence of submucosal antibodies which gives the buccal mucosa a normal resistance [7].

Tuberculosis of the tongue is a rarely described entity with a rate of 0.1% to 1.0% [7]. The tongue does not contain any significant lymphoid tissue for which tubercular mycobacterium has a great affinity [8] So there is a relative inhibition of the growth of mycobacteria in the tongue in particular [8] The most common presenting symptoms of lingual tuberculosis were pain on deglutition, followed by burning sensation and otalgia [8]. Oral lesions typically are ulcers on the lateral border, tip and dorsum of the tongue. The ulcer is usually formed by breakdown of tubercles and with undermined edges and erythematous base [7]. Tubercular ulcers are usually more irregular and indurated than punched out lesions of carcinoma. Oral tuberculous lesions may also occur on the gingiva, floor of mouth, palate, lips and buccal mucosa [7-9].

The differential diagnosis of an oral tuberculous ulcer includes aphthous ulcers, traumatic ulcers, syphilitic ulcers and malignancy, including primary squamous cell carcinoma, lymphoma and metastases [9]. The most likely clinical diagnosis is that of squamous cell carcinoma, where biopsy is mandatory and an oral ulcer is only considered tuberculous when the biopsy reveals an epithelioid granulomatous lesion, as our case revealed. Other oro-facial granulomatous conditions are sarcoidosis, Crohn’s disease, the deep mycoses, cat-scratch disease, foreign-body reactions, tertiary syphilis and Melkersson-Rosenthal syndrome [10].

A detailed clinical history and examination are important to make up the diagnosis, and ancillary laboratory tests with radiography are helpful; although the histopathologic biopsy remains the golden standard for confirmation of the diagnosis.
The confirmation of tuberculosis is by the presence of acid-fast bacilli in Z-N stain and culture of the tubercular bacilli. Sputum culture and chest radiograph are mandatory, since oral tuberculosis is almost always secondary to pulmonary tuberculosis. Self-inoculation may take place from infected sputum or haematogenous seeding [12]. Malignancy per se and treatment modalities used for malignancy, both lead to immune suppression, and can thus, unmask pulmonary TB [13]. Coexistence of carcinoma tongue with pulmonary TB is rare and, in India, where most of the adult population is already infected with Mycobacterium tuberculosis, a diagnosis of reactivation/reinfection pulmonary TBs should be kept in mind while evaluating any patient with malignancy [2,13].

The present case differs from previously reported cases because the patient presented with an ulcerated lesion which was clinically suspected as malignancy. This case also highlights the need to take deep biopsies from the edge of ulcerated lesions in order to avoid misdiagnosis/underdiagnosis due to non-representative biopsies which could be interpreted only as non-specific chronic inflammation.

CONCLUSIONS

In patients presenting with non-healing tongue ulcers, the possibility of tuberculous glossitis should be included in the differential diagnosis and a representative and “deep tissue” biopsy should be performed so that malignancy is not missed.

REFERENCES