Tuberculosis of Oral Cavity: A Series of One Primary and Three Secondary Cases

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Received: 21 October 2011 / Accepted: 19 January 2012 / Published online: 22 March 2012
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Abstract Tuberculosis is one of the most common diseases in India resulting in significant mortality and morbidity. The disease has varied presentations. The clinical spectrum ranges from usual primary tuberculosis of the lungs to the very rare of oral cavity. We are reporting four cases of tuberculosis of oral cavity one of which is primary and three are secondary. All the patients were adult males. The primary tuberculosis case was sputum and biopsy negative for acid fast bacilli. All the secondary oral tuberculosis cases showed cavitary lesions in lungs and were sputum positive for acid fast bacilli however the biopsies were negative for the same.

Keywords Tuberculosis · Tongue · Buccal mucosa · Primary · Secondary

Introduction

Tuberculosis is endemic in India. It was a disease more frequent in developing and underdeveloped countries however with the advent of AIDS, the global pattern evolved over time and it assumed significance in hitherto comparatively unaffected affluent societies [1, 2]. Global

burden of tuberculosis is 8 million new cases every year and 3 million deaths. It is estimated that, in developing countries 30–60% of the population is infected by tuberculosis [3, 4]. With emerging menace of Extensively drug resistant tuberculosis in addition to already significant global burden owing to Multi drug resistant tuberculosis, future of tuberculosis as a global problem appears to be far from over.

Tuberculosis is one of the most prevalent diseases in India. It is a chronic granulomatous disease caused by *Mycobacterium tuberculosis*. Less frequently the pathogen is mycobacteria other than tuberculosis causing tuberculosis like illness.

Tuberculosis displays rather wide clinical spectrum. In majority of cases primary infection is acquired by bacilli impregnated droplet inhalation and hence the sites of primary infection are the lungs in majority of cases of tuberculosis. However primary site may also be skin, bone, Gastrointestinal tract and Genitourinary system but much more often these site are affected secondary to the primary focus in the lungs [5].

Primary tuberculosis in the oral cavity however remains rare even in a country like India where the disease is rampant. So far cases of primary tuberculosis of the tongue are published as an anecdotal case reports because of extreme rarity [6]. Tubercular foci in oral cavity secondary to primary tuberculosis of the lung is again uncommon if not rare [7]. The oral presentation of tuberculosis has an incidence of only 0.1–0.4% [8].

Tuberculosis of oral cavity even as secondary form is uncommon [7].

Here we are reporting one case of primary tuberculosis of the tongue and three cases of secondary tuberculosis of oral cavity (two of tongue and one of oral mucosa near the angle of mouth).

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Case #1 (Primary Tuberculosis of Tongue)

A 65-year-old-male presented with pain in the base of tongue and difficulty in swallowing. There was history of loss of appetite and weight (Fig. 1).

On examination there was an ulcerated lesion on the posterior one third of tongue with irregular margins. The base of ulcer was indurated and firm but not fixed to the underlying muscles. Chest X ray was normal and sputum was negative for AFB.

A provisional diagnosis of malignancy was kept on the basis of clinical examination.

General physical examination revealed mild pallor but no significant lymphadenopathy of the neck was seen.

The patient had Hemoglobin, 9 mg/dl, Total leukocyte count was LC, 6800/cu mm and differential leukocyte count showed mild lymphocytosis. ESR, bleeding time, clotting time, blood urea, creatinine, sugar along with chest X ray and ECG were carried out which did not reveal any abnormality.

Biopsy of the ulcerated lesion showed epitheloid cell granulomas and Langhan type of giant cells along with surrounding lymphoplasmacytic infiltrate. Stain for acid fast bacilli on the histopathology slides was negative. A diagnosis of features consistent with tuberculosis was made.

Case# 2 (Secondary Tuberculosis of Tongue)

A 54 years old male presented in the ENT OPD with complaint of pain and ulceration over the right lateral border of tongue for the last 20 days. There was no history of fever, cough, chest pain, loss of weight and loss of appetite (Fig. 2).

On examination, the size of ulcer was 2×1.5 cm covered with white stuff. Margins of ulcer were irregular. On palpation, base of the ulcer was indurated and firm but not fixed. The lesion was tender.

Fig. 1 Primary tuberculosis of tongue. Ulceration over posterior tongue

Fig. 2 Secondary tuberculosis of tongue. Ulcerations at lateral margins of tongue

Clinically provisional diagnosis of malignancy was kept.

General physical examination revealed pallor but no significant lymphadenopathy of the neck was seen.

Biopsy of the tongue was planned and accordingly routine relevant investigations in the form of Hb, TLC, DLC, ESR, BT, CT, Urea, Creatinine, Sugar along with chest X ray and ECG were carried out.

X rays chest revealed cavitory lesion in the left upper zone of chest. Subsequently three consecutive sputum samples for AFB were sent and all of them were positive for AFB.

A punch biopsy of the ulcer from the margin was taken. Microscopic examination showed stratified squamous epithelium, acanthosis and parakeratosis. The subepithelial areas showed fibrocollagenous tissue and inflammatory infiltrate along with a single ill formed epitheloid cell granuloma and scattered Langhan's type of giant cells. No deeper tissue was included in the biopsy (Fig. 3).

The diagnosis of secondary tuberculosis of the tongue secondary to primary sputum positive lung tuberculosis was made.

Fig. 3 Microphotograph, high power of H&E stained section. Representative picture of epitheloid cell granuloma

Case # 3 (Secondary Tuberculosis of Tongue)

A 70-years-old-male presented with the history of pain and ulcer on the left lateral border of the tongue and excessive salivation. The ulcer was smaller to start with but was gradually increasing in size. There was no history of cough, fever and chest pain. However there was history of loss of appetite and weight along with night sweats.

On examination, the ulcer was 3 × 2.5 cms and covered with white stuff. It had irregular margins and lateral border of tongue was eaten up. Base of the ulcer was indurated, tender and firm in consistency.

A provisional diagnosis of malignancy was made.

Routine and relevant investigations in the form of blood and chest X ray were asked for. Punch biopsy from the margin of the ulcer was taken and sent for histopathological examination.

The X ray chest revealed cavitory lesion on the right upper zone and sputum was positive for AFB. Histopathology report was consistent with tuberculosis of tongue.

Case # 4 (Secondary Tuberculosis of Buccal Mucosa Near the Angle of Mouth)

A 35-years-old-male presented with history of ulcer over the buccal mucosa near the angle of mouth. The ulcer had irregular margins and base was irregular and firm in consistency.

Relevant routine investigations were normal. X ray chest revealed cavitory lesion and sputum was positive for AFB. Histopathology report revealed features consistent with tuberculosis of oral mucosa.

Discussion

The pathogenesis of oral TB usually is self inoculation with infected sputum, resulting from the constant coughing up of bacteria that seed themselves in the oral tissue along their line of discharge through the mouth or it may result from hematogenous dissemination [2].

Although there are many patients with pulmonary tuberculosis, lingual form is not frequently met with despite of the fact that tongue is frequently exposed to heavy concentration of tubercular bacilli from the sputum of patients with pulmonary tuberculosis. This local resistance of the tongue may be due to the stratified squamous epithelium, regular cleaning by saliva, presence of skeletal muscle and paucity of lymphoid tissue in anterior part of tongue [9].

In addition role of chronic trauma cannot be underestimated which breach the first defense against infection in

the form of stratified squamous epithelium [10]. Secondary tuberculosis of the oral cavity is commoner than the primary. The percentage of patients with primary pulmonary tuberculosis developing infection of oral cavity varies from 0.1 to 0.5%. Secondary tuberculosis most commonly affects the tongue, followed by the palate, the lips, the buccal mucosa, and the gingival [5]. The salivary glands, tonsils and uvula also are involved frequently [1]. In addition secondary lesions of the mandibular ridge (alveolar mucosa) are also reported which are extremely rare [11].

Similar to the other body parts clinical manifestations of oral tuberculosis are varied. In the oral cavity, it usually manifests as non healing ulcers but may also occur as nodules, granulomata or fissures, as tuberculous osteomyelitis of the jaws [10]. Oral TB ulcers are usually single rather than multiple; they have an indurated, irregular and undermined margin with a necrotic base. The ulcer may be initially painless but may become painful with the passage of time [12]. Other manifestations of oral tuberculosis may include nodular lesion or cold abscess [6, 13].

Three forms of oral tuberculosis have been described: acute miliary, chronic ulcerative, and lupus vulgaris [14]. Overwhelming majority (about 93%) of the oral lesions are ulcers and approximately half of which affect the tongue. [15].

All the cases in our series are chronic ulcerative. The ulcers were on the lateral margins of tongue and buccal mucosa near the angle of mouth. The latter is again very rare.

Primary tuberculosis of the oral cavity is extremely rare and the published literature is only in the form of case reports [5, 6, 16]. There appear to be geographical variations regarding which age group is affected by primary tuberculosis. In western literature, it is mentioned that primarily oral tuberculosis affects children and adolescents and is often associated with enlarged cervical lymph nodes [17]. A few case reports from India also fall into younger age group [6, 16]. Tuberculosis of oral cavity can be seen in adults but primary tuberculosis of lip and uvula amongst pediatric age group has been reported occasionally [16]. Weng et al. [17] reported a series of 20 cases from Taiwan wherein 55% of the patients were older than 50 years and the most common location was buccal mucosa and/or vestibule (5 cases), followed by the alveolar mucosa (4), palate (2), lip (2), and tongue (1 case).

There is a similarity in age distribution between what Weng et al. reported and our cases however it is remarkable that only one out of 20 had tongue involvement in the former while there are three out of four cases of tuberculosis of the tongue in the present study.

Rarely, direct inoculation may result in primary oral tuberculosis. The site most commonly affected is the

gingiva where primary TB appears as a diffuse erythematous patch or as diffuse gingival enlargement [18].

Primary tuberculosis of the oral cavity has been reported in HIV infected immune deficient cases as an indicator of HIV infection; however most of the published literature pertains to cases with intact immune status similar to the case under discussion [5, 19].

Secondary oral TB may occur in all age groups; however, middle-aged and older people more likely will have oral manifestations of the disease, which are almost always painful [20].

Secondary tuberculosis of the tongue manifests in two ways: one with shallow, oval, indolent, painful ulcers with overhung margins and the other with a circumscribed interstitial tuberculoma. The pain is because of intact nerve endings and both types of lesions occur in advanced untreated cases of tuberculosis. Other tuberculosis lesions—diffuse glossitis or fissures—have also been reported but they are exceedingly rare [9]. Although the dorsal surface is more commonly involved, involvement of the ventral surface has also been described [9].

Differential Diagnosis and Diagnosis

The differential diagnosis of tuberculous ulcers includes traumatic ulcers, aphthous ulcers, Vincent's angina, actinomyces and carcinoma [12].

The diagnosis of tuberculosis is made by biopsy which should show epithelioid cell granulomas with Langhan's type of giant cells. In biopsy, the deeper tissue must be included as the granulomas are seen in the deeper dermis. The superficial biopsies only show hyperplasia of stratified squamous epithelium. The demonstration of acid fast bacilli in the biopsy material further confirms the diagnosis.

Alternatively the tissue may be subjected to molecular diagnostic methods like polymerase chain reaction or even culture. While the former is sensitive and specific, it is also technically demanding and expensive. The culture is still considered to be the gold standard for diagnosis with the additional advantage that precise drug sensitivity of cultured bacilli can be carried out. However it is time consuming and takes weeks.

Thus tuberculosis should be born in mind as a differential diagnosis of oral ulcers. It is important since clinically the lesion may simulate malignancy particularly when the patient happens to be among the middle or older age group.

It is noteworthy that in all the cases in the present series the common theme was a chronic painful ulcerated lesion and clinical diagnosis of malignancy which is much more common than tuberculosis of the tongue. In none of the cases a clinical diagnosis of tuberculosis was suspected

initially. The biopsies however revealed epithelioid cell granulomas with Langhan's type of giant cells. All the three cases of secondary oral tuberculosis showed cavitary lesions in the lungs and sputum samples were positive for acid fast bacilli. The case of primary tuberculosis of the tongue, however, the diagnosis was made purely on the basis of histopathological report.

We propose that initial therapy for tuberculosis ought to be started as soon as the biopsy report comes in consistent with tuberculosis. All of our cases responded very well to the standard regimen (DOTS) of for 6 months. The lesions had healed completely.

The purpose of this series of case reports is to highlight the rare clinical presentation of tuberculosis and bring to the attention a differential diagnosis of tuberculosis while dealing with oral ulcers. It is particularly relevant in a country like India with one of the highest tuberculosis burden in the world.

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