

Primary Lingual Tuberculosis Mimicking Malignancy: A Report Of Two Cases And Review Of Literature

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Citation

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Abstract

Every year, approximately 2 million people in India develop tuberculosis, accounting for one fourth of the world's new tuberculosis cases. However primary tuberculosis of tongue is very rare. Here we report two cases of primary lingual tuberculosis in a 56-year-old female suffering from idiopathic thrombocytopenic purpura and in a 32-year-old male without any known predisposing factor.

INTRODUCTION

Every year, approximately 2 million people in India develop tuberculosis, accounting for one fourth of the world's new tuberculosis cases [1]. Incidence of tuberculosis in India is 168/100 000 population/ year and prevalence is 312/ 100 000 population/ year [2]. However oral manifestation of tuberculosis is rare with an incidence of 1.4% of total tuberculosis cases [3]. Floor of mouth, soft palate, gingiva, lips, and hard palate can be involved, however tongue [tip, dorsum, lateral borders, and base] and palate are the most common sites of involvement for oral lesions [4]. Primary tuberculosis lesion of the tongue is very rare, the secondary may occur in those having pulmonary tuberculosis affecting 0.05-0.5% of elderly tuberculosis patients [4,5,6]. Primary tuberculosis of upper aerodigestive tract has also been reported in larynx, soft palate and tonsil [3]. To best of our knowledge only 10 cases of primary lingual tuberculosis have been reported [3, 4]. Clinical presentations of tongue tuberculosis vary and different pattern of gross lesions have been described.

Here we report the eleventh case of lingual tuberculosis in a 56 year old female with known idiopathic thrombocytopenia (ITP) on glucocorticoids for 1 year due to persistence of platelet count below 50,000/ μ l. Localized manifestation of opportunistic infection secondary to use of steroid is also rare. Another rare occurrence of primary lingual tuberculosis in a 32 year-old male without any predisposing factors is also reported here.

CASE REPORTS

CASE 1

A 56 year old female residing in Saharanpur, Utter Pradesh, India, presented in the outpatient department of department of otolaryngology of All India Institute of Medical Sciences with complaints of increased salivation, pain and ulcer in the tongue. The pain was low grade, dull aching and nonradiating type. The patient was diagnosed to have ITP [7] one year back and was on oral Wysolone 20mg alternate day since then. There was no history of cough, fever and hemoptysis or weight loss. The patient belonged to low socioeconomic strata and was an occasional tobacco chewer since last 20 years.

On examination her general built and heights were normal. A 2x3 cm ulcer was seen in the left lateral border of tongue with elevated margins and dirty necrotic base. Surrounding induration was identified. Adjacent teeth were normal. Nasopharynx and oropharynx were also clinically normal. No cervical or axillary lymph nodes were palpable. A provisional diagnosis of malignancy was considered.

Laboratory investigations revealed hemoglobin 11.3gm% with a microcytic hypochromic picture, TLC 16,000/cumm, DLC- N₇₅, L₁₈, M₆, E₁, platelet 55, 000/ μ l, erythrocyte sedimentation rate 35mm at 1 hour, plasma glucose 92 mg% (fasting) and 116 mg% (post-prandial). Other biochemical investigations were within normal limits. Mantoux test performed with PPD antigen showed negative result after 72 hours. Rapid card test for HIV was negative. Chest skiagram AP view examined was within normal limits with no hilar

and mediastinal lymphadenopathy.

Biopsy taken from the ulcer margin showed mucosal ulceration with presence of necrotizing epithelioid cell granulomas among the underlying skeletal muscle fibers with Langhans giant cells and lymphocytic infiltrate [Fig. 1]. Overlying lingual mucosa was hypertrophied [Fig. 2]. Zeihl Neelsen stain performed showed numerous acid fast bacilli [Fig. 3]. A histological diagnosis of tuberculosis was given. The diagnosis was further confirmed by polymerase chain reaction. On further workup no evidence of tuberculosis was identified elsewhere.

Figure 1

Figure 1: Hyperplastic lingual mucosa seen with underlying granulation tissue and granuloma. H & E, X40.

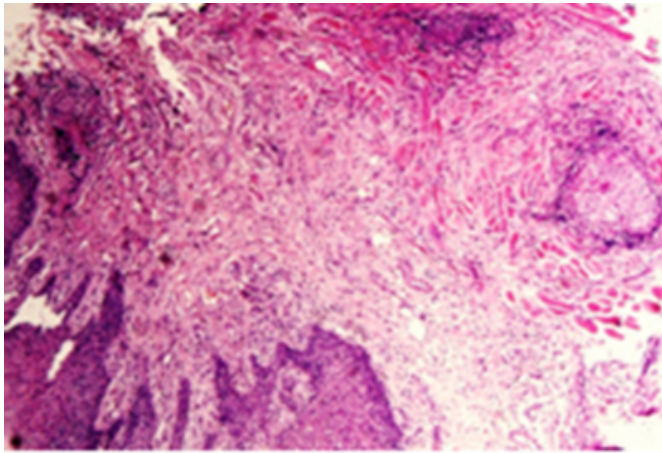


Figure 2

Figure 2: Epithelioid granuloma. Skeletal muscle fibers are also identified. H & E, X100.

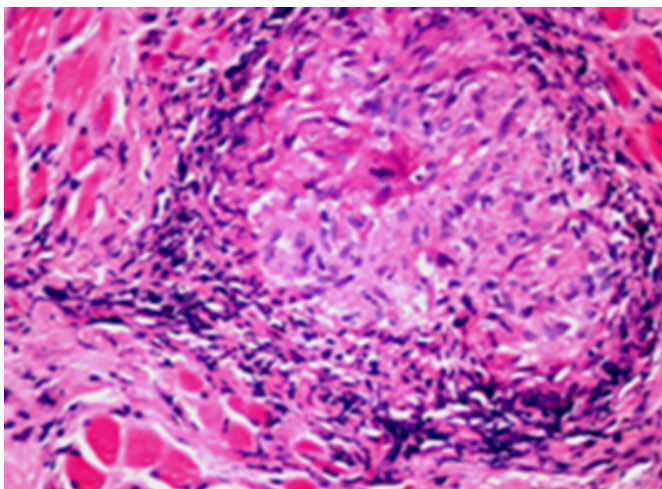
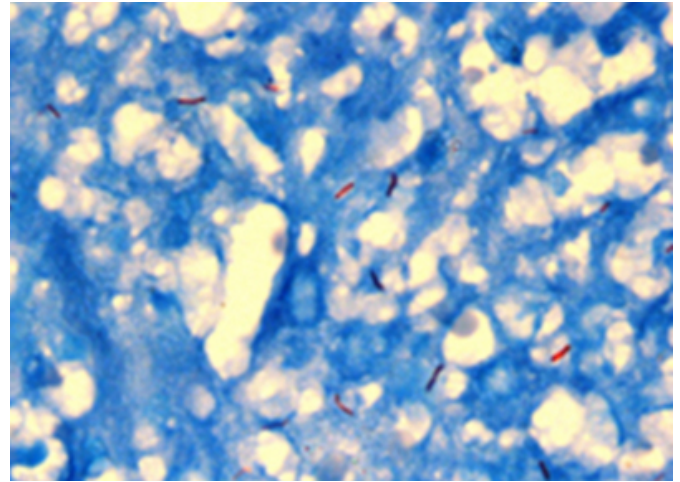


Figure 3

Figure 3: Slender beaded tubercle bacilli inside the granulomas. ZN stain, X 1000.



The patient was placed on an antitubercular therapy with gradual tapering and then withdrawal of glucocorticoids. Repeated platelet counts were within safe limits.

CASE 2

A 32 year old male residing in Delhi, India, presented in the outpatient department of department of otolaryngology of All India Institute of Medical Sciences with complaints of increasing pain and ulcer in the tongue. The pain was dull aching in type and increased while chewing foods. No history of cough, fever, and hemoptysis or weight loss was found. A similar low socioeconomic status and history of tobacco chewing was found since last 9 years. The patient used to wash tooth with coal ashes.

On examination general built was thin along with mild pallor. No lymphadenopathy was found. On auscultation bilateral chest was within normal limits. Cardiovascular system was normal. On local examination a 5.2 x3 cm ulceroproliferative lesion was seen in the left lateral border of anterior third of tongue. Margins of the lesion were elevated and base was necrotic. Clinically a diagnosis of malignancy was made.

Laboratory investigations revealed a hemoglobin of 9.8 gm% with a microcytic hypochromic picture, TLC 9,000/cumm, DLC- N₉₂, L₆, M₂, E, platelet 300, 000/ µl, erythrocyte sedimentation rate 22 mm at 1 hour, plasma glucose 108 mg% (fasting) and 134 mg% (post-prandial). Other biochemical investigations were within normal limits. Chest AP view examined was within normal limits.

Initial biopsy performed showed mucosal ulceration with

underneath inflammatory granulation tissue. No evidence of granuloma was found. A full course of standard oral antibiotic therapy was not successful. Due to clinical suspicion of malignancy a repeat, deeper biopsy was performed. This time well formed necrotizing granulomas with giant cells and inflammatory cells were identified among the muscle fibers. A stain for acid-fast bacillus was positive. The diagnosis was further confirmed by polymerase chain reaction.

DISCUSSION

In India tuberculosis is a major health hazard with a mortality rate of 30 deaths/100000 population/year, even after the National tuberculosis control programme [NTCP] has brought down the prevalence rate significantly [2]. Oral tuberculosis is common in 20- 40 years age group with a male to female ratio of 4: 1 and poor socio-economic class. In a large series by Komet et al [1965] the most common presenting symptoms of lingual tuberculosis were pain on deglutition, followed by burning sensation and otalgia [8]. The most common site of lingual tuberculosis described in the literature are lateral border followed by tip, posterior third, middle third, ventral surface and anterior surface [9]. Different types of gross presentation are seen like: painful shallow tubercular ulcer, tuberculoma, tubercular fissure, tubercular papilloma and tubercular cold abscess [4, 5, 9, 10]. The ulcer is usually formed by breakdown of tubercles and usually has undermined edge [9]. Many forms of presentations may mimic malignancy as, nodular [11] and ulcers not typical of tuberculosis. Tubercular ulcers are usually more irregular than punched out lesions of carcinoma. [5, 12]. In our cases the tubercular ulcer had elevated margins. In atypical cases only mode of diagnosis is either detection of tissue acid fast bacillus, detection of tubercular antigens by polymerase chain reaction or culture isolation.

In primary oral tuberculosis the organisms are directly inoculated on the oral mucous membrane of a person who has not been previously infected. The role of trauma is controversial, as the stratified squamous epithelium of the oral cavity normally resists direct penetration by tubercle bacilli. In the secondary type, oral tuberculosis usually coexists with pulmonary disease. Self inoculation may take place from infected sputum or hematogenous seeding [13]. In our cases in absence of source of tuberculosis in any part of body the bacilli may have been inoculated in tongue from aerosols. Immunosuppression due to prolonged use of steroids for treatment of ITP may have been the predisposing

factor in first case. However such factor was not present in the second case. We cannot rule out a role of traumatic inoculation of tubercular bacilli in this case as the patient used to wash tooth with crude coal ashes. Differential diagnosis of oral ulcers includes traumatic ulcer, aphthous ulcer, actinomycosis, syphilitic ulcer, Wagener granuloma and carcinoma. Diagnosis is made by identification of a caseating granuloma on biopsy. Deeper biopsies are advocated for ulcers of tongue because superficial biopsies may not be true representative of the lesion; furthermore, multiple biopsies may be needed [10].

Treatment followed in our country for tuberculosis is direct observation antitubercular therapy [DOTS]. Surgery is not required in these cases and prognosis is favorable. In patients who require immunosuppression due to any cause the possibility of opportunistic infections should be included in the differential diagnosis.

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