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Case Report

PRIMARY ISOLATED TUBERCULOUS ORAL VESTIBULE – A CASE REPORT

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ABSTRACT
Tuberculosis of the oral cavity is rare and when present is usually secondary to pulmonary tuberculosis. We present a case of a 45-year-old male patient who complained of a non-healing ulcer in the right oral vestibular area. Local examination revealed a white patch over the right vestibule. Biopsy was consistent with the diagnosis of primary tuberculosis vestibule. The patient had no evidence of tuberculosis anywhere else in the body. Antituberculous treatment was curative. As this condition is very rare, it is often over looked until and unless proved otherwise. The relevant literature is reviewed.

Key Words: *Vestibule, Antituberculous Treatment, Primary Tuberculosis, Oral Tuberculosis*

INTRODUCTION
Tuberculosis is a global health problem, with eight million people newly infected annually and three million people dying from its complications. Its incidence in developing countries is increasing probably due to associated poor hygienic conditions (Tanwar *et al.*, 2012; Wang *et al.*, 2009). The primary route of transmission is the respiratory tract. Primary oral tuberculous lesions are much rarer probably due to early diagnosis and treatment elsewhere in the body.
The purpose of this article is to emphasise the importance of early diagnosis especially when oral lesions are not associated with any apparent systemic infection.

CASES
A 45 year old male belonging to lower socio-economic status presented with a painful oral ulcer in the right vestibule for past 6 months. The lesion did not heal even with medications.
There was no history of dental trauma, cough with expectoration, fever, weight loss or night sweats and his appetite was normal. There was no history suggestive of tuberculosis elsewhere in the body or in the family. He was non-diabetic and non- hypertensive. There was a significant history of bidi smoking for the last 30 years with an average of one and half bundle per day.
On examination, his general condition and vital signs including blood pressure, pulse and respiratory rate were within normal limits. There was no cervical lymphadenopathy. Systemic review was unremarkable. Local examination revealed a tender ulcer in the right vestibular region against the lower pre-molar tooth. The teeth and rest of the oral cavity was normal with poor oral hygiene. The ulcer was solitary and ill-defined with dimensions of 1.5 cm x 2.5 cm. The surface of the ulcer was shiny and no bleeding or pus discharge was observed.
Routine investigations revealed haemoglobin of 13.5 gm/dL, WBC count of 9000 cells/mm3, Polymorphs 80 %, Lymphocytes 20%, and ESR 17 mm in 1st Hr. ELISA for HIV and RPR for Syphilis were non-reactive. PPD test was equivocal. Sputum examination for Acid fast bacilli (AFB) was negative. Chest X-ray was normal. A punch biopsy was taken. The subsequent histopathological examination showed acanthotic stratified squamous epithelium and connective tissue with the presence of multiple epithelioid cell granulomas, Langhan's giant cells and tiny areas of caseation (Figure 1). Acid-fast bacilli (AFB) staining was positive by modified Ziehl-Neelsen staining (Figure 2). Thus, a diagnosis of isolated primary gingival tuberculosis was made.
The patient was given antituberculous treatment and there was complete resolution of the disease and during follow-up there has been no recurrence, suggesting a successful outcome.

DISCUSSION

Tuberculosis is a chronic infectious granulomatous disease and is very common in India and Southeast Asia, where the prevalence rate is about four for every 1000 people and the incidence rate of the disease is of two percent (Tanwar *et al.*, 2012; Rodrigues *et al.*, 2007; Carnelio and Rodrigues, 2002).

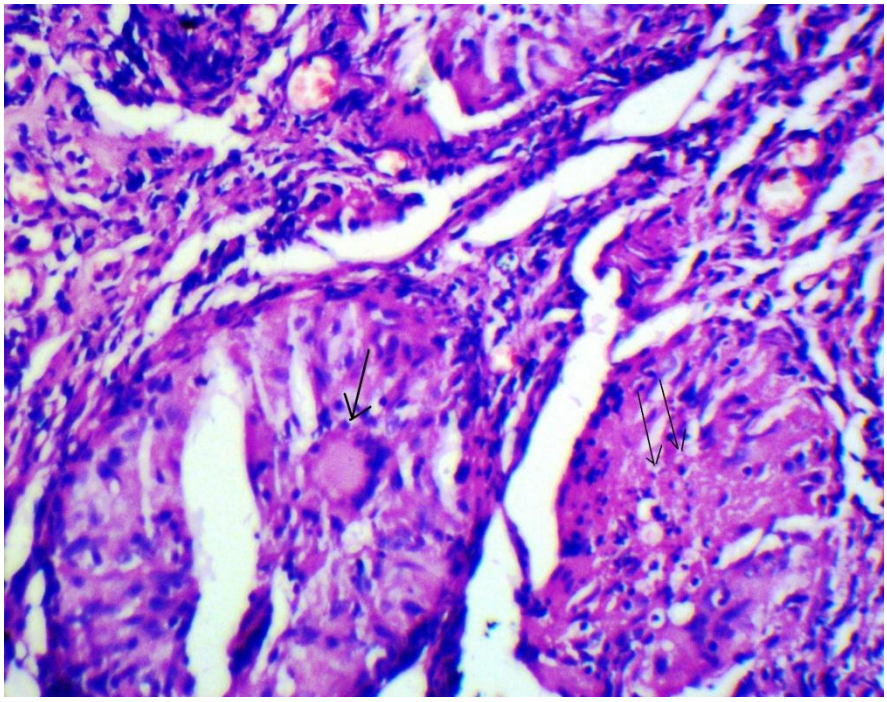

Figure 1: Photomicrograph showing acanthotic squamous epithelium, granulomas comprising of Epithelioid cells, Langhan's giant cells (↓) and tiny areas of caseation (↓↓) [H&E X 400]

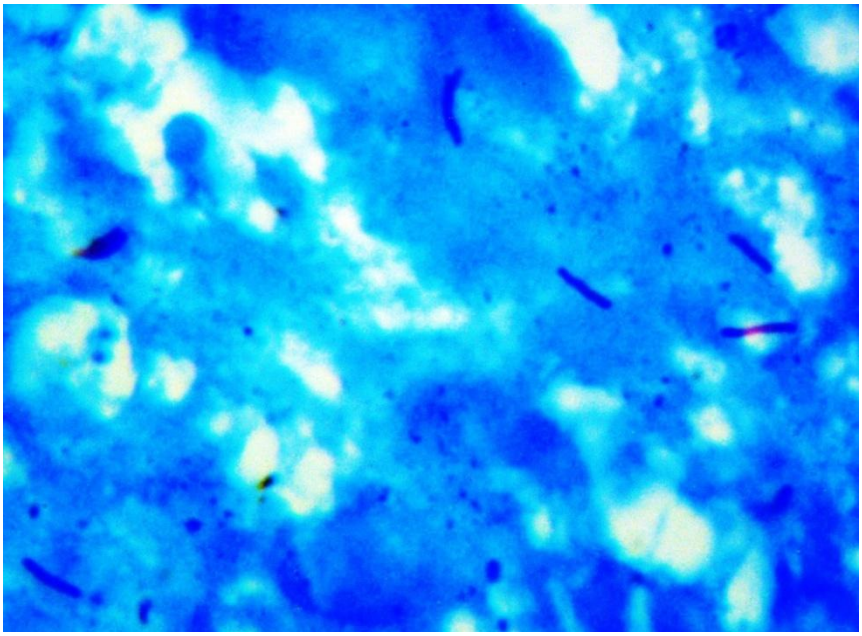

Figure 2: Photomicrograph Showing Acid Fast bacilli. [Modified Ziehl-Neelsen Stain X 1000]

Case Report

Oral tuberculosis is caused by *Mycobacterium tuberculosis* and less frequently by *Mycobacterium bovis*, and is usually associated with tuberculosis of the oropharynx, lungs, lymph nodes and milliary form. Both primary and secondary types have been reported but all are associated either with Candidiasis, HIV infection, pulmonary tuberculosis or cervical lymphadenopathy (Carnelio and Rodrigues, 2002). Primary isolated vestibular tuberculosis is extremely rare and there have been only a handful of cases documented in the literature (Tanwar *et al.,* 2012; Rodrigues *et al.,* 2007). Tongue and gingiva are the most common sites of infection in patients with oral tuberculosis, followed by that of tooth sockets, soft palate, floor of mouth, lips and buccal mucosa (Carnelio and Rodrigues, 2002; Ito *et al.,* 2005). Oral lesions can also present as nodules, fissures, plaques, vesicles, tuberculomas or granulomas. They may be single or multiple, painful or painless and usually appear as irregular, well circumscribed ulcer with surrounding erythema without induration and satellite lesions. They progressively extend from the gingival margin to the depths of the adjacent vestibule and are often associated with enlarged cervical lymph nodes (Tanwar *et al.,* 2012).

The pathogenesis is usually self inoculation with infected sputum, but haematogenous spread is also known. Direct inoculation commonly involves the gingiva, dental sockets and buccal folds. It is believed that an intact squamous epithelium of the oral mucosa is relatively resistant to penetration of tuberculous bacilli and that saliva has some inhibitory effect on mycobacteria. For primary oral lesions (where there is no evidence of old or current pulmonary tuberculosis), the mycobacterium may directly inoculate oral mucosa following minor injury as a result of trauma, inflammatory conditions, or tooth extraction (Wang *et al.,* 2009). If the amount of mycobacterium in an ulcerating oral tuberculosis lesion is high enough, there is the possibility that more lesions may occur and also there may be dissemination to other individuals via the saliva. Other predisposing factors include poor oral hygiene, leucoplakia and greater prevalence of AIDS (Mignogna *et al.,* 2000; Gupta *et al.,* 1998). Vestibular tuberculosis should be differentiated from traumatic ulcers, aphthous ulcers, actinomycosis, candidiasis, syphilitic ulcers, Wegener's granulomatosis, sarcoidosis and malignancies including lymphoma. A biopsy, as in our patient, is necessary to confirm the diagnosis. Chest X-Ray and Mantoux skin test are mandatory to rule out systemic tuberculosis. The treatment of oral tuberculosis is by anti tuberculous drugs.

The diagnosis of oral or facial tuberculosis can be quite difficult, mainly because of a lack of definite signs and symptoms. Cases showing AFB positivity may decline when more epithelioid cell granulomas are observed (Tanwar *et al.,* 2012). The diagnosis of primary lesion in our patient was made on biopsy because the clinical features of the oral lesions were non-specific and chest radiographs and sputum examination, including Ziehl–Neelsen staining for pulmonary involvement were negative.

Conclusion

Even with decreasing incidence tuberculosis still remains a major health problem, especially in India. Efforts to eradicate it have been hindered by poverty, lack of healthcare access, drug resistance, immuno-suppressed populations and global migration.

Whether it is primary and secondary oral tuberculosis, early detection, diagnosis, and treatment are of the utmost importance. Effective management requires prompt recognition using a combination of clinical, radiographic, microbiological, and histopathologic correlation with initiation of appropriate multidrug therapy.

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