

Atypical presentation of tuberculosis

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Abstract

Developing countries such as India face an intense situation with communicable diseases such as tuberculosis (TB). It is an inflammatory infectious granulomatous disease commonly affecting the lungs. Extrapulmonary TB presentations have been documented, with the orofacial region also being involved. TB usually presents as an ulcer in the oral cavity with tongue and lip being the favored sites. TB presenting as a lip swelling is unusual even if it is a secondary lesion with a primary pulmonary involvement. We present a lip swelling and discuss the differential diagnosis. Investigation of ultrasonography revealed a vascular lesion which was disproved, suggesting an increase in vascularity even in diseases such as TB. We present a case of a lip swelling which led to the diagnosis of recurrent pulmonary TB which the patient was unaware of. Findings of histopathology revealing the absence of bacilli are also discussed. Monetary constraints frequently cause a neglect of dangerous diseases such as TB.

Keywords: Histopathology, lip, swelling, tuberculosis

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INTRODUCTION

Tuberculosis (TB) is a chronic granulomatous infectious disease. It still remains a major health concern, especially in developing countries. It primarily affects the lungs, with the head and neck being a rare site of extrapulmonary involvement. Areas involved in the head and neck include cervicofacial lymph nodes, oral cavity, pharynx and larynx. The cervical lymph nodes are the common area of presentation. A diagnosis of head-and-neck TB helps in establishing a more serious pulmonary connection of the infection. The manuscript throws light on the differential diagnosis of an asymptomatic lip swelling and the use of ultrasonography with interesting negative findings of histopathology.

CASE REPORT

A 55-year-old male reported with a chief complaint of a swelling of the lower lip which had been present for the past 4 months [Figure 1]. History revealed that he was apparently symptomless until 4 months ago when he developed this swelling gradually and was treated by a general physician with antibiotics for the same. The swelling did not regress which made the patient seek our consultation, and he currently only complained of the lip swelling. Upon eliciting medical history, we were informed that he was treated for pulmonary TB 4 years ago when he completed his antitubercular treatment (ATT) Directly observed treatment, short course (DOTS) and was certified free of disease. He had a history of smoking

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Figure 1: Extraoral picture showing diffuse enlargement of the lip on the day of presentation

2–5 bidis (country-made cigarettes) in a day for the past 40 years. He also occasionally chewed areca nut and tobacco.

On clinical examination, the lower lip was diffusely swollen measuring roughly 4 cm × 3 cm, but was not involving tissues beyond the normal lip boundaries. No obvious surface changes were observed. On palpation, the consistency was soft with no paresthesia or pain and no discharge of pus or blood. Keeping in mind the age of the patient and the presentation of the lip swelling, an allergy as in the case of a contact allergy or angioneurotic edema was thought of but later ruled out based on the duration and history of the patient where no inciting contact agent could be isolated. With the same history of no associated trauma or inciting event, the diagnosis of a possible foreign-body reaction was ruled out.^[1]

Differential diagnosis

A blood vessel anomaly was considered. Arteriovenous malformation is a disease with a broad age range. The start of the disease is insidious with the possibility of surface changes in the skin and mucosa. The lesions are usually soft and compressible. The common sites involved are the lip and cheek (80%).^[2]

Cheilitis granulomatosa could present as an isolated lip swelling. It does not have a definite age, sex and gender predilection. When associated with facial palsy and tongue fissuring, a diagnosis of Melkersson–Rosenthal syndrome may be thought of, but both these signs were not present in our case.^[1,3]

Orofacial granulomatosis are a group of diseases based on exclusion and was retained as an important differential diagnosis in the present case. It includes diseases such

as Crohn's disease, sarcoidosis and TB. Crohn's disease patients could sometimes develop a diffuse swelling of the lips and face. The patients are usually aged above 40–50 years. Coexisting cobblestone appearance of the mucosa with persistent deep linear ulcerations may be seen sometimes.^[3] However, the patient did not complain of any associated gastrointestinal symptoms which are invariably associated with Crohn's disease, in which a lip swelling could be the only presentation.

Sarcoidosis could also present as an enlarged lip, but the lack of associated systemic symptoms helped us rule out this entity. TB is very common in South-East Asian countries and should rank high on the list of differentials. The typical presentation is usually an undermined ulcer on the tip of the tongue and less commonly on the lip. In our case, the sole presentation was the lip swelling, but keeping in mind the previous history of treated TB, we retained it as an important differential.^[4]

Deep fungal infections could also affect the lip but the swelling and associated symptoms are invariably progressive which was in contrast to our case where it was static once it reached the present size 1 year ago.

A tumor of the salivary gland could have been a possibility but the diffuse nature of the swelling ruled it out as minor salivary gland tumors are rather well circumscribed.^[1]

Idiopathic macrocheilia was also thought of in case all advised investigations turned out negative.

Diagnosis

After obtaining necessary informed consent, a complete hemogram was performed but no significant deviation from the normal ranges was observed. Due to the financial constraints of the patient, an ultrasonography of the lip was performed which revealed a diffuse area of soft-tissue thickening measuring 40 mm × 18 mm approximately. On color Doppler, the lesion showed both arterial and venous waveforms, with PSV ranging from 15 cm to 25 cm/s which was suggestive of a vascular lesion [Figure 2]. Based on these findings, an aspiration was performed to confirm the vascular nature of the lesion but was found to be negative. This was in contradiction to the ultrasonography findings. We went ahead and performed an incisional biopsy in a hospital setup in anticipation of an inadvertent bleed, with tissue taken from the intraoral labial aspect of the swelling. The biopsy specimen was fixed in 10% formalin to be processed for H and E staining. The stained section showed dense inflammatory infiltrate chiefly composed of lymphocytes in the connective tissue. A few foci of central

eosinophilic areas resembling caseation necrosis were seen, surrounded by clusters of epithelioid cells and histiocytes attempting to form giant cells. Many endothelial-lined blood vessels were also seen in the connective tissue. Based on clinicopathological correlation, a diagnosis of a tuberculous granulomatous lesion was arrived at [Figures 3–6].

Management

Keeping in mind our final diagnosis of tuberculous lip swelling and as per the regional guidelines for the management of a patient with TB, he was referred to the Regional TB Treatment Centre where the following investigations were performed: chest radiograph and CB-NAAT sputum examination for acid-fast bacilli (AFB) with TB-polymerase chain reaction being reserved as a second line of investigation if and when required. The chest radiograph revealed involvement

with consolidation of the upper lobes [Figure 7]. Sputum examination was positive for AFB in the CB-NAAT. The findings did not necessitate further investigations, and he was started with Category II ATT [Table 1]. Two-month post-ATT showed a significant resolution of the swelling [Figure 8]. Keeping in mind all the features and results of investigations, we concluded that this case

Table 1: Treatment schedule given to the patient

Category II	
Treatment Schedule	Drugs
Month 1, 2	Isoniazid Rifampicin Ethambutol
Month 3, 4, 5 and 6	Isoniazid Rifampicin

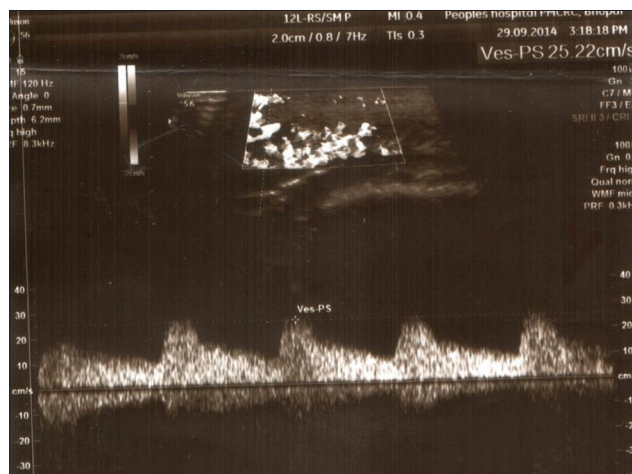


Figure 2: Ultrasonogram of the lower lip showing a high peak systolic volume

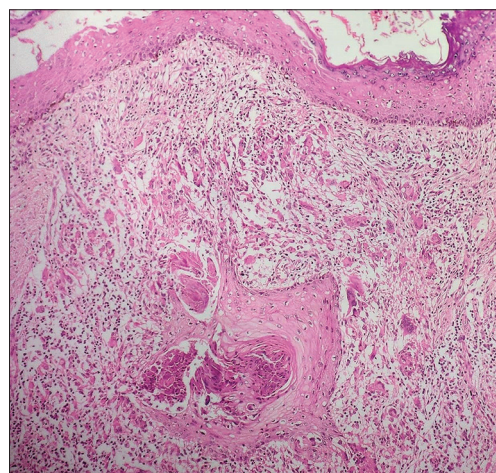


Figure 3: Histopathological picture at $\times 10$ revealing parakeratinized stratified squamous epithelium with underlying fibrovascular connective tissue with evidence of focal granulomas

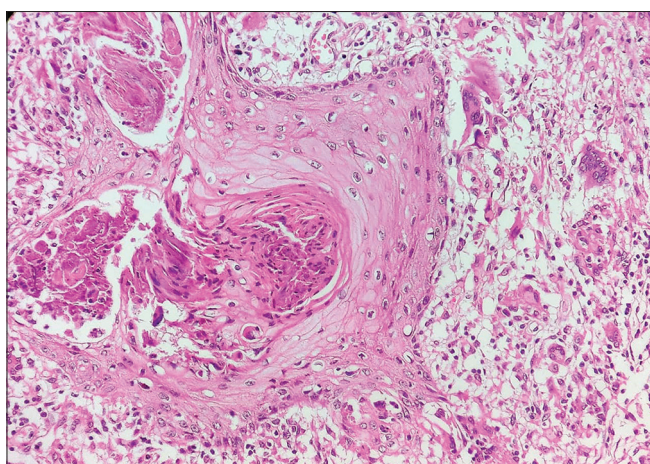


Figure 4: Histopathological picture at $\times 20$ showing focal granulomatous area with central caseation necrosis surrounded by epithelioid cells and chronic inflammatory cell infiltrate and endothelial-lined blood vessels with a few multinucleated giant cells

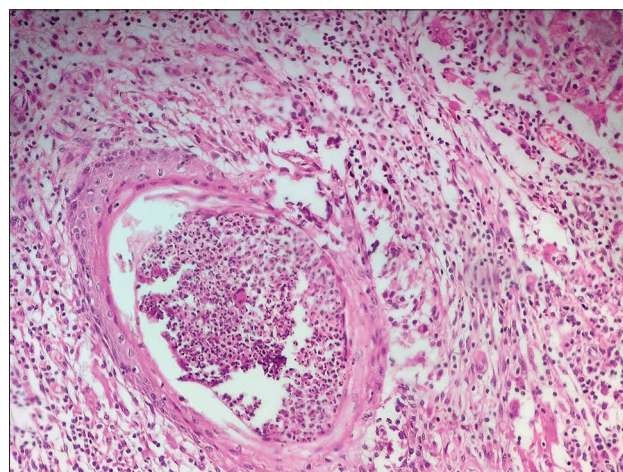


Figure 5: Histopathologic picture at $\times 20$ showing focal granulomatous area with central caseation necrosis with mixed inflammatory cell infiltrate surrounded by epithelioid cells along with endothelial-lined blood vessels and chronic inflammatory infiltrate with lymphocytes predominantly is evident

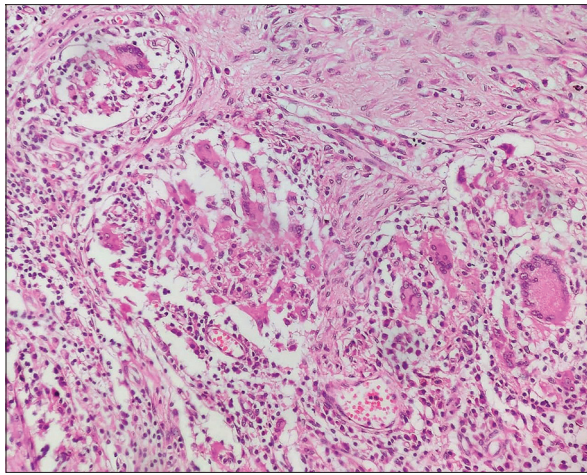


Figure 6: Histopathologic picture at x20 showing focal granulomatous areas with epithelioid cells and multinucleate giant cells

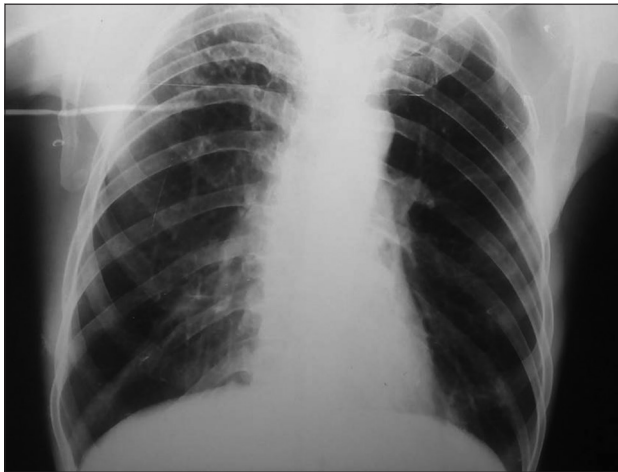


Figure 7: Chest radiograph revealing the involvement of bilateral upper lobes



Figure 8: Posttreatment photograph after 2 months of Category II antitubercular treatment showing resolution of the lip swelling

was a reactivated case of pulmonary TB with coexisting oral findings.

DISCUSSION

The incidence of TB is extremely high in developing countries. The World Health organization has estimated 2 million deaths/year due to TB, while roughly 36 million people may die of the disease by 2020.^[1] TB is primarily a respiratory disease although other organs of the body may also be affected simultaneously or individually. The presentation of oral TB is of two types, a primary type which is common in younger individuals and with no pulmonary involvement, and a secondary type which is common in older individuals with up to 96% of patients having evidence of lung lesions. In children, it invariably presents as lymphadenitis of the orofacial region.^[4] TB usually presents as an ulcer in the oral cavity with tongue and lip being the favored sites. TB presenting as a lip swelling is unusual even if it is a secondary lesion with a primary pulmonary involvement. In the oral cavity, the spectrum of lesions is extremely diverse, ranging from a painless or painful ulcer, erosion, erythematous patch, nodule and fissure either presenting singly or in multiples.^[5,6] The presence of salivary enzymes and tissue antibodies provides saliva with an inherent cleansing action and property to fight infections. When an infection does occur, the etiopathogenesis is presumed to be due to a loss of barrier or break in the continuity of epithelium. The loss of barrier could be due to the conditions including inflammatory lesions, inadequate oral hygiene or a simple tooth extraction.^[7,8] The ultrasonography finding of arterial and venous waveforms correlated with the histopathological finding of numerous endothelial-lined blood vessels in the connective tissue, suggesting probable increased vasculature in the labial tuberculous lesions requiring further investigations.

A biopsy is extremely crucial to distinguish TB lesions from neoplastic lesions as the treatment greatly varies.^[8] It seems logical to keep in mind the diagnosis of secondary TB when a patient presents with a diffuse swelling of a lip. In a country like India, even though the appearance of the lesion was nonspecific, TB of the lip should rank high in the differentials of nonspecific diffuse swelling of the lip in elderly patients with a history of completely treated pulmonary TB.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given his consent for his images and other clinical information to be reported in the journal. The patient understands that name and initials will not be published, and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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