

Sarcoidosis of Tongue

Raghuram Pitti, Ramesh Kumar, Saravana S Selvam, Eswar Nagaraj

ABSTRACT

Sarcoidosis is a rare, acquired systemic granulomatous disease, with unknown etiology affecting multiple organs and tissue. A 40-year-old male with the previous history of pulmonary sarcoidosis was screened with the clinical presentation of erythematous and nodular lesion over the tongue. Incisional biopsies from the tongue revealed the presence of 'non-caseating epithelioid cell granulomas with Langerhans type multinucleated giant cells' histopathologically. The 24 hours urine calcium examination revealed increased calcium level of 538 mg suggestive of oral sarcoidosis. The patient is periodically reviewed to study the course of the disease and identify the systemic progression at the illness.

Keywords: Granulomatous disease, Pulmonary sarcoidosis infection, Dorsal surface of tongue.

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INTRODUCTION

Sarcoidosis is a multisystemic granulomatous disease of unknown etiology. Lymph node, lungs, eyes are commonly affected.¹ The respiratory system is most commonly affected with approximately 90% of cases with pulmonary findings.² Parotid gland and the cervical lymph nodes are most frequently affected in head and neck region.³ Oral involvement in sarcoidosis is relatively very rare. The first oral lesion of sarcoidosis was reported in 1942 by Schroff. There are only seven cases of sarcoidosis of tongue reported till date in literatures.² It has high incidence in women than men.

CASE REPORT

A 40-year-old male patient was reported to the Department of Oral Medicine and Radiology with the chief complaint of missing teeth. History revealed that the patient had pulmonary sarcoidosis infection and was undergoing treatment since 2001. History revealed that the patient was diagnosed for sarcoidosis and confirmed to radiograph, sputum culture and sensitivity test, elevated ACE levels and unresponsive to antituberculosis therapy (ATT).

Intraoral examination revealed multilobulated granulomatous growth in the dorsal surface of tongue with irregular margins which mimicked that of median rhomboid glossitis (Fig. 1).

Angiotensin converting enzyme (ACE), serum calcium, 24 hours urine calcium were advised which revealed an elevated 24 hours urine calcium of 538.0 mg.

Incisional biopsies were taken from the tongue which revealed 'Langerhans type of multinucleated giant cells' (Fig. 2). Microbiological test in sputum were negative. The histopathologic findings was suggestive of sarcoidosis. Correlating the history, clinical and investigatory findings the diagnosis of oral sarcoidosis was arrived at.

DISCUSSION

Oral lesions occurs in chronic multisystem sarcoidosis and it may be solitary multiple or a part of generalized disease.¹ In some cases oral involvement may be the first or the only manifestation of the disease.⁴



Fig. 1: Intraoral picture of granulomatous growth on the dorsal surface of tongue

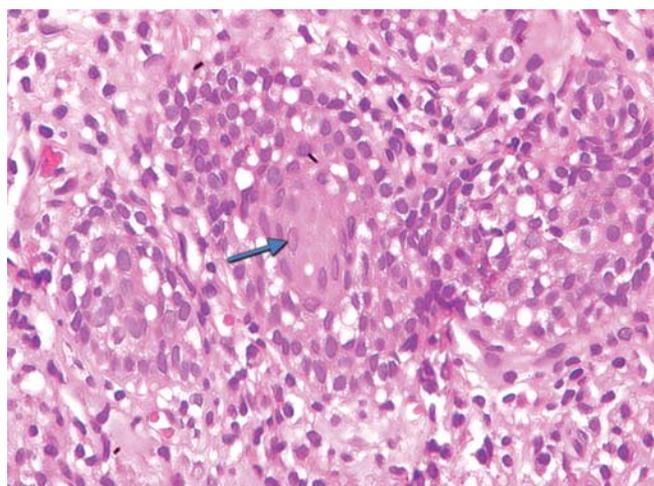


Fig. 2: Multinucleated Langerhans giant cells

While the etiology of sarcoidosis remains to be elucidated, there is consensus that the pathogenesis of the disease is the result of activated mononuclear phagocytes and oligoclonal CD4+ T cells driving a polarized TH1 immune response to yet unknown tissue antigens. This immune response is characterized by increased production of interferon (IFN).

Clinically, sarcoidosis may present acutely or in a sub-acute or chronic fashion. Distinct presentations of sarcoidosis are associated with different clinical courses, with approximately 50% of patients undergoing remission, usually within 2 to 3 years.⁵ The other 50% of patients have persistent, generally progressive disease requiring treatment to mitigate the consequences of unremitting inflammation and subsequent fibrosis.

There are no specific tests for sarcoidosis. Diagnosis of sarcoidosis is mainly based on clinical characteristics as well as chest X-ray and exclusion of other noncaseating granulomas forming conditions. S-ACE is positive in 80% cases of sarcoidosis and may be false negative in some cases with other systemic diseases.⁶

In our case report although atypical a well circumscribed depapillated lesion was evident over the dorsal surface of tongue and granulomatous in nature. The multinodular growth on the tongue differed from previous reported cases which usually presented with sessile swellings.

In addition chest radiograph revealed linear fibrotic strands and hilar vascular shadow, a common pulmonary presentation of sarcoidosis and TB. The presence of TB was ruled out through sputum and sensitivity tests showing absence of AFB. Also the raise in S-ACE to 62.4 u/l may be attributed to active form ongoing T-cell and macrophage inflammation resulting in increased ACE confirming sarcoidosis.⁷

CONCLUSION

Although very rare, oral lesions of sarcoidosis may be the first presenting symptom. This multisystemic disorder can never be completely cured. Glucocorticoids, antimalarial

drugs and immunosuppressive agents form the standard regimen for sarcoidosis treatment. They are associated with potential toxicities. Some authors may suggest surgical excision of oral soft tissue or lesion. At present we suggest a periodic follow-up of patients which may be helpful in evaluation of the course and progression of the disease.

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