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Sarcoidosis of the tongue: A case report

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Abstract

A case of sarcoidosis involving the tongue is described in a 48-year-old Japanese man. A definite diagnosis of sarcoidosis was made for the clinical lesion and pathological examinations. Sarcoidosis is a multisystem granulomatous disease that may affect any organ. Sarcoidosis of the tongue is particularly rare.

Keywords: Sarcoidosis; Tongue; Corticosteroids

1. Introduction

Sarcoidosis is a systemic granulomatous disease of unknown etiology. The organs most frequently affected are the lungs, lymph nodes, eyes, and skin. In the head and neck region, the parotids glands and cervical lymph nodes are most often involved in sarcoidosis. Sarcoidosis of the tongue is extremely rare. To our knowledge, there are only seven cases of sarcoidosis involving the tongue reported to date [1–7]. Here, we report in detail a new case.

2. Case report

A 48-year-old Japanese man was referred with a 2-month history of slowly increasing swelling and induration of the tongue. He had difficulty in eating and weight loss. On examination, there was indurated swelling on the left side of the tongue, however, the overlying mucosa was intact (Fig. 1). On palpation, a firm, submucosal mass was felt. This tongue mass did not have pain. Mobility of the tongue was slightly reduced. No cervical lymph nodes were palpable, and the salivary glands were not enlarged. The remainder of the head and neck examination was normal. Magnetic resonance imaging showed an enhancing mass at the left side of the tongue (Fig. 2). Under general anesthesia, the lesion of the tongue was subtotally excised. Histological examination revealed non-caseating epithelioid cell granulomas with Langerhans type multinucleated giant cells (Fig. 3). There was no microscopic evidence of acid-fast bacilli fungi, or foreign bodies.

In view of the histological diagnosis further examinations were carried out. The chest X-ray showed bilateral hilar lymphadenopathy (Fig. 4A). ^{67}Ga scintigraphy disclosed abnormal isotope uptake in the bilateral hilus of the lung (Fig. 4B). However, pulmonary function tests were normal. His serum angiotensin converting enzyme (26.9 IU/l; reference range: 8.3–21.4 IU/l) and lysozyme (27.8 μg/ml; reference range: 5.0–10.2 μg/ml) levels were elevated. Other laboratory investigations, including full blood count, immunoglobulins, serum calcium, and liver function tests were all normal (Table 1).

A diagnosis of sarcoidosis was made on the based of these findings, and further investigations instituted. There was involvement of his eyes and spleen. Computed tomography of the abdomen showed multiple nodules in the spleen (Fig. 5). The tuberculin skin reaction was positive.

Treatment was begun with administration of prednisone 60 mg daily with subsequent reduction to 10 mg. After 1 year of steroid therapy, his tongue has remained normal in size and his ocular disease and pulmonary lesions showed a marked improvement.

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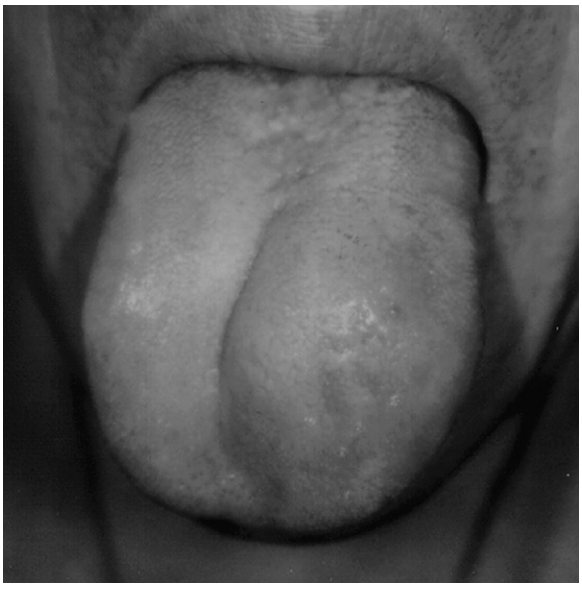

Fig. 1. Indurated swelling lesion on the left side of the tongue with intact mucosa.

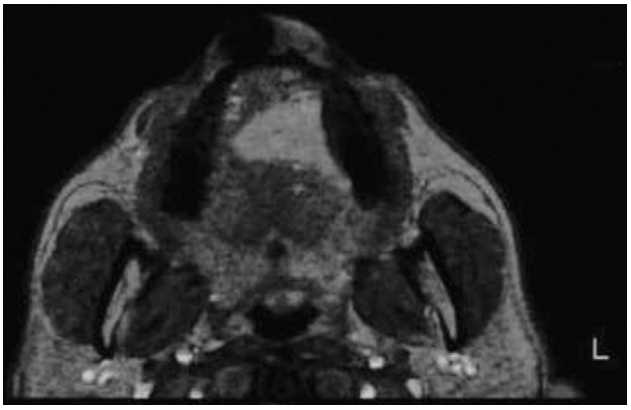

Fig. 2. Gadolinium-enhanced, T1-weighted MRI showing an enhancing mass at the left side of the tongue.

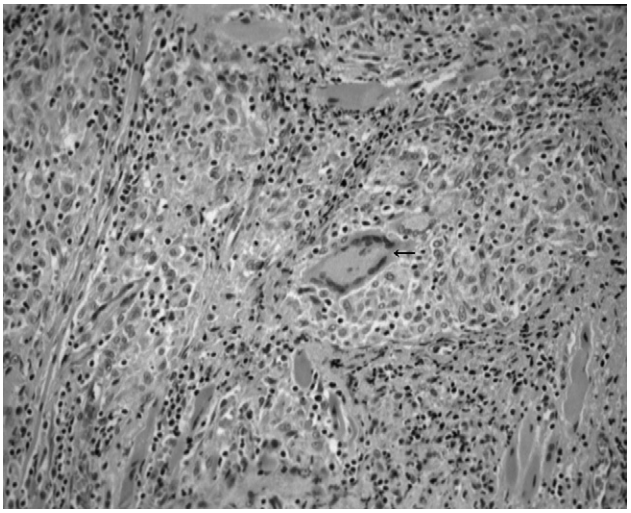

Fig. 3. Non-caseating epithelioid cell granulomas with Langerhans type multinucleated giant cells (arrow). (Hematoxylin–eosin stain; ×100).

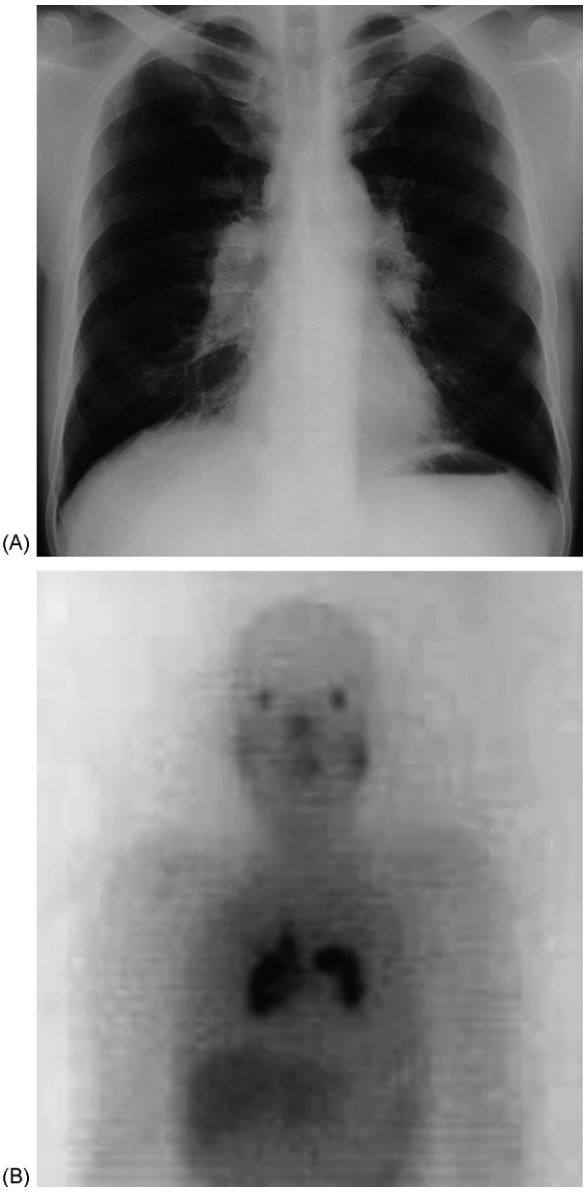

Fig. 4. (A) The chest film showing bilateral enlargement of the hilar lymph nodes. (B) ^{67}Ga scintigraphy showing abnormal isotope uptake in the bilateral hilus of the lung.

Table 1
Labo data

		reference range
WBC	4.24	3.50 ~ 9.00 x 10^3/ μl
RBC	4.50	4.50 ~ 5.50 x 10^6/ μl
Plate	381	140 ~ 440 x 10^3/ μl
IgG	1597	872 ~ 1815 mg/dl
IgA	283	95 ~ 405 mg/dl
IgM	97	31 ~ 190 mg/dl
Ca	9.7	8.7 ~ 10.3 mg/dl
AST	22	13 ~ 33 U/l
ALT	20	6 ~ 30 U/l

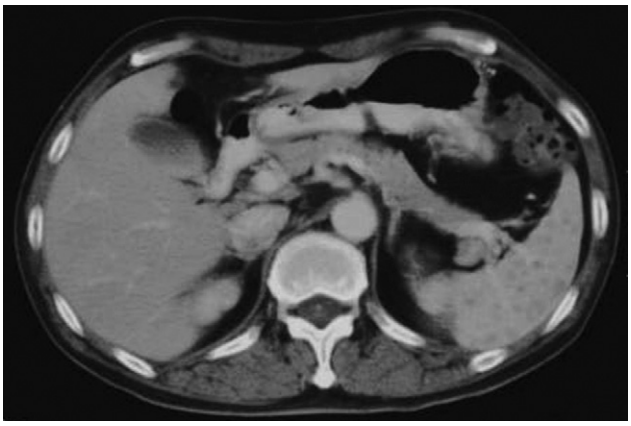

Fig. 5. Computed tomography of the abdomen showing multiple nodules in the spleen.

3. Discussion

The tongue is rarely involved in sarcoidosis; to our knowledge, only seven cases have been reported [1–7]. The tongue alone was involved in three cases [1,4,6] and pulmonary lesions were found in four cases [2,3,5,7]. Spleen involvement was only found in our case. According to literature, swelling and induration of the tongue was found in three cases [1,6,7], an erosive and popular lesion was found one case [5], and nodular lesion was found in three cases [2–4]. Our case had swollen and indurated tongue.

The diagnosis is established when clinical findings are supported by histological evidence of typical granulomas. Differential diagnosis includes foreign body granulomas and orofacial granulomatosis, such as oral Crohn's disease, granulomatous chelitis, and Melkersson's syndrome. Tuberculosis, syphilis, actinomycosis, and other infections should be included in the differential diagnosis because they may make a sarcoid-like tissue response. Carcinoma should also be considered in the differential diagnosis.

The levels of serum angiotensin converting enzyme and lysozyme are assumed to show the state of sarcoidosis. In this case, lungs, eyes, and spleen were actually affected.

Corticosteroids seem to be the only drug capable of curing or stopping the progression of sarcoidosis. Most sarcoidosis is treated by corticosteroids, but mild asymptomatic cases did not require systemic therapy. Surgical excision was used to remove fibrous bands of tongue after treatment using corticosteroids [1]. Our case was treated with systemic therapy using corticosteroids after surgical excision of lesion on the tongue.

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