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Kaposi's Sarcoma of the Tongue Associated with Median Rhomboid Glossitis in a Non-AIDS Patient. A Case Report

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Abstract. *Kaposi's sarcoma (KS) of the tongue is extremely rare in immunocompetent patients. We report a case of KS of the tongue associated with a median rhomboid glossitis. The main clinical, pathological and immunohistochemical features allowed the differential diagnosis.*

Kaposi's sarcoma (KS) is a multicentric vascular neoplasm, which may be classified as classic, endemic, post transplant and epidemic or AIDS-associated KS (1,2). All clinical forms of KS are characterized by a similar morphological picture and are strongly related to herpes virus 8, that may be considered as an infectious cofactor responsible for all types of KS (3). Several vascular lesions can mimic KS, including angiosarcoma, hemangioendothelioma, pyogenic granuloma and kaposiform hemangioendothelioma (1,2,4,5). Oral KS is a very rare disease in immunocompetent patients (6,7), whereas it is more frequent in immunocompromised individuals. Exceptional examples of KS of the tongue have been reported in the literature (8-10).

We describe a case of KS of the tongue, arising in an immunocompetent patient that formerly exhibited only a median rhomboid glossitis. The unusual association, as well as the exceptional localization, prompted us to report this case.

Case Report

A 67-year-old Caucasian man was referred as an outpatient to the Head and Neck Pathology Department of the 2nd University of Naples, Italy. At clinical examination, a glossitis with rhomboid appearance was observed. A small incisional biopsy was performed and the diagnosis of median rhomboid glossitis with extensive chronic inflammatory infiltrate was made. Six months later, oral examination showed a red-white nodular lesion (midline region of the tongue), with partially ulcerated surface, measuring 1.5 cm (Figure 1). No clinical evidence of transplantation, immunosuppression, AIDS, blood transfusion or body irradiation was revealed by the clinical history of the patient.

On the basis of these data, a pyogenic granuloma was suspected. An excisional biopsy was carried out. The tissue sample was fixed in formalin, then routinely processed and embedded in paraffin. The sections were stained with haematoxylin-eosin, Van Gieson, PAS, Giemsa and Gomori's silver stain.

Additional sections were cut and subjected to immunohistochemical studies. The streptavidin-biotin-peroxidase complex method (DAKO, Copenhagen, Denmark) was performed using heat-induced antigen retrieval. As primary antisera, the following antibodies (DAKO) were employed: S-100 protein, vimentin, CD 31, CD 34, alpha-smooth-muscle actin and Factor VIII-related antigen (FVIII-RAg).

Histological examination displayed an ulcerated, infiltrative lesion, prevalently composed of intersecting fascicles of moderately atypical spindle-shaped cells, showing ill-defined cytoplasm and plump oval nuclei, generally characterized by small nucleoli (Figure 2). These cells often presented intracellular lumen containing eosinophilic globules and erythrocytes.

Between the fascicles, clefts and slit-like vascular spaces were observed. Mitotic activity was high (more than 20 mitosis per 10 high-power fields). Occasional atypical mitotic features were detected in spindle cells. Small clusters of plasma cells and lymphocytes were intermingled among neoplastic cells.

Immunohistochemically, the spindle cells were strongly positive for vimentin, CD 31 (Figure 3) and CD 34 (Figure 4). No staining for FVIII-RAg, actin and S-100 protein was detected in neoplastic cells.

In view of these morphological and immunohistochemical findings, the diagnosis of KS of the tongue was made.

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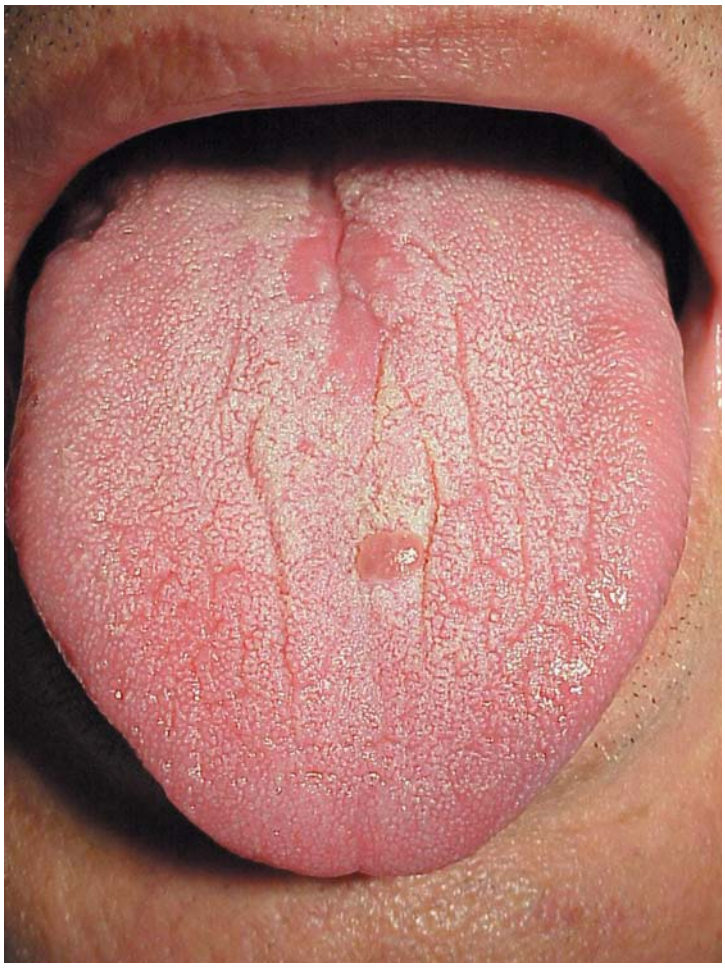

Figure 1. *Red-white nodular lesion of the tongue associated with median rhomboid glossitis.*

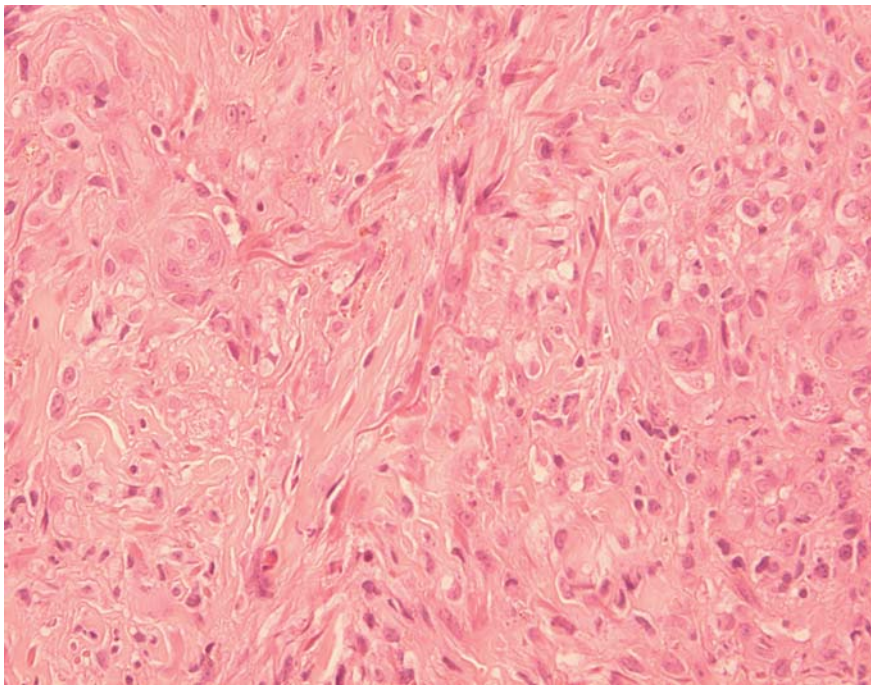

Figure 2. *Irregular vascular spaces, spindle cells and red blood cells "entrapped" (haematoxylin-eosin,400X).*

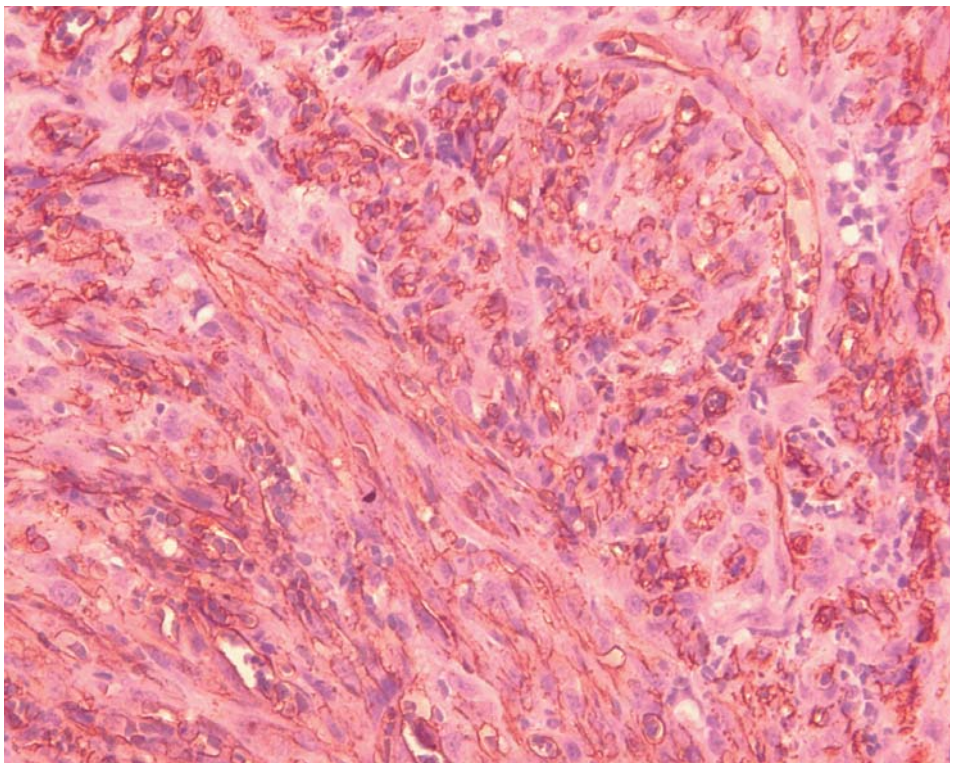

Figure 3. *CD 31 staining in Kaposi's sarcoma tissue (ABC, 400X).*

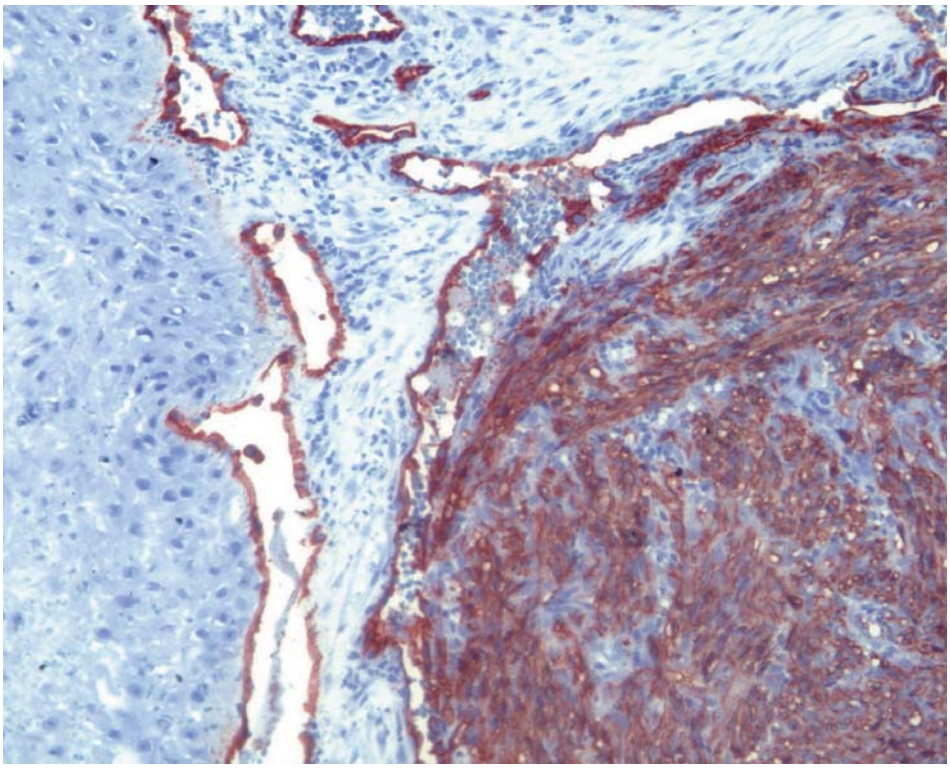

Figure 4. *CD 34 staining identifiable only in Kaposi's sarcoma tissue. Overlying stratified epithelium appears negative (ABC, 400x).*

Discussion

As regards the differential diagnosis, some lesions might be kept in mind, particularly in relation to the differentiation of KS from pyogenic granuloma (PG) and Kaposi-like hemangioendothelioma (KLH). Indeed, macroscopic and clinical aspects of the present case were considered by us to be suggestive of PG.

Nevertheless, some morphological features of this case are usually not detected in PG, such as fascicle of spindle cells characterized by intracellular lumen containing erythrocytes and eosinophilic globules, clefts and slit-like vascular spaces. The immunohistochemical profile (CD 34+, CD31+, FVIII-RAg-) was that usually expressed by KS. In addition, various morphological characteristics of PG were lacking in the present case: epithelial collarette, lobular architecture, myxoid stroma and granulocytic infiltrate (4,11-13).

Moreover, KLH very rarely occurs in adults and displays some histopathological features not identifiable in the present case, such as the infiltrative lobular pattern, vascular microthrombi, glomeruloid proliferation and low mitotic rate (14,15). In addition, small clusters of plasma cells, not identifiable in KLH, were detected.

Therefore, the morphological and immunohistochemical data were considered compatible with the diagnosis of Kaposi's sarcoma.

The relationship between median rhomboid glossitis and KS of the tongue appears to be intriguing, although this association has never been reported in the literature. This type of glossitis is considered a developmental abnormality caused by the persistence of an embryonic midline tongue structure, also known as the tuberculum impar (12,13). This condition is often associated with chronic infection by *Candida albicans*, suggesting that some immunological local defects may occur and make this area more susceptible to this infection (12,13).

It is tempting to hypothesize that this local defect might also increase the susceptibility to the infection by herpes virus 8, which is involved in the pathogenesis of KS.

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