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## Case report

### A case of AIDS-associated oral Kaposi's sarcoma of the tongue

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#### ABSTRACT

Kaposi's sarcoma (KS) is one of the most common diseases seen in patients presenting with acquired immunodeficiency syndrome (AIDS); however, it is rare in Japan. We herein report a case of AIDS-associated KS of the tongue, which was initially misdiagnosed as recurrent hemangioma according to the initial histopathological diagnosis. The patient is a 42-year-old male who had been suffering from a painful vascular neoplasm-like mass on the dorsum of the tongue. The patient did not complain of any other distinct symptoms and a debulking operation was planned based on the clinical diagnosis of hemangioma. However, preoperative blood tests revealed the presence of syphilis and the human immunodeficiency virus and the patient was therefore diagnosed to have full-blown AIDS. Therefore, the patient's oral lesion was then instead suspected to be oral KS (OKS). A histopathological examination of the tongue biopsy specimen showed the typical findings of KS. Combination active antiretroviral therapy (cART) combined with liposomal doxorubicin was administered and the patient achieved a complete remission (CR). In conclusion, clinicians including oral surgeons, should take OKS into account in the diagnosis of vascular neoplasm-like masses of the tongue in adults since this complication may occur as a result of AIDS.

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## 1. Introduction

Kaposi's sarcoma (KS) is a well known vascular tumor first described by Moriz Kaposi in 1872 [1]. This angioproliferative disorder, which is characterized by the proliferation of spindle-shaped cells, neoangiogenesis, inflammation, and edema [2], is categorized as an intermediate neoplasm due to the absence of conventional features of malignancy [3].

Four major forms of KS have so far been identified: classic, African endemic, immunosuppression-associated or transplant-associated, and AIDS-associated [4]. The histopathologic and immunohistochemical features are similar in all of the clinical forms. In addition, human herpesvirus 8 (HHV-8) DNA is found in almost all cases of KS and in all of the clinical forms [5].

AIDS-associated KS (AIDS-KS), the most aggressive form of the disease, is found in human immunodeficiency virus-1 (HIV-1)

infected individuals and it is particularly frequent in homosexual and bisexual men. Although the relative risk of acquiring KS for AIDS patients is >10,000 fold [6], the incidence of KS has been reduced with the advent of combination active antiretroviral therapy (cART) [7].

Head and neck involvement in AIDS-KS is common [8]. Oral lesions represent the first sign of KS in 22% of HIV-positive individuals, and ultimately, 71% of these patients will develop AIDS-associated oral KS (AIDS-OKS) [2,9]. AIDS-OKS has been reported to occur most frequently on the hard palate, followed by the gingiva and the tongue [5,10–12].

In this report, we present a rare case of AIDS-OKS which occurred on the tongue that led to the discovery of AIDS in a Japanese man.

## 2. Case report

A 42-year-old Japanese male was referred to our hospital for evaluation of a painful mass on the dorsal surface of the tongue that had slowly increased in size. The patient underwent a resection of a neoplastic lesion on the dorsal surface of the tongue at another hospital approximately 5 months before the initial examination at our hospital. The resected specimen was histopathologically diagnosed at the previous hospital as hemangioma. After the operation, the patient suffered from throat pain and began to suspect a recurrence

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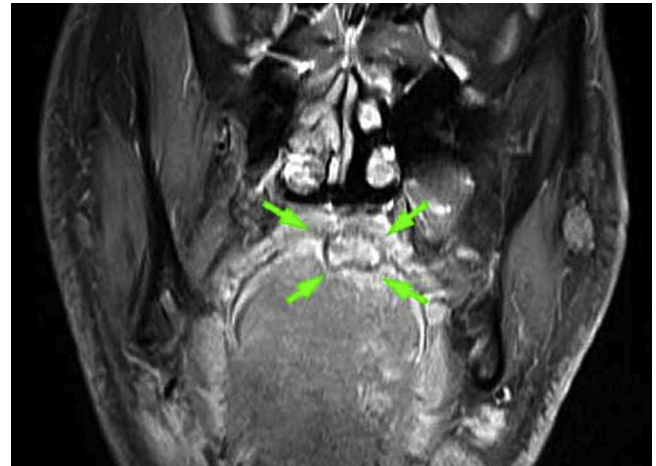


**Fig. 1.** The appearance of the tongue lesion at the initial examination. A blue to purplish, nodular, exophytic mass measuring 2 cm × 2 cm was found to occupy the left side of the dorsal surface of the tongue. (For interpretation of the references to color in this figure legend, the reader is referred to the web version of the article.)

of the neoplasm. The patient's past history included diet-controlled diabetes mellitus diagnosed at 30 years of age and hepatitis B contracted at 33 years of age.

No facial asymmetries, cervical lymphadenopathies, or other distinct extraoral findings were detected. Upon intraoral examination, a blue to purplish, exophytic, and vascular neoplasm-like mass measuring 2 cm × 2 cm was found to occupy the left side of the dorsal surface of the tongue (Fig. 1). The findings of the remaining oral cavity and oropharynx were within normal limits. MRI scans of the head and neck revealed only a small superficial lesion on the dorsal surface of the tongue without regional metastasis or infiltration into the deep tissue (Fig. 2). Laboratory data at the first examination are shown in Table 1. All of the hemogram data showed subtle decreases, and the biochemistry data were within normal limits except subtle increase in C-reactive protein.

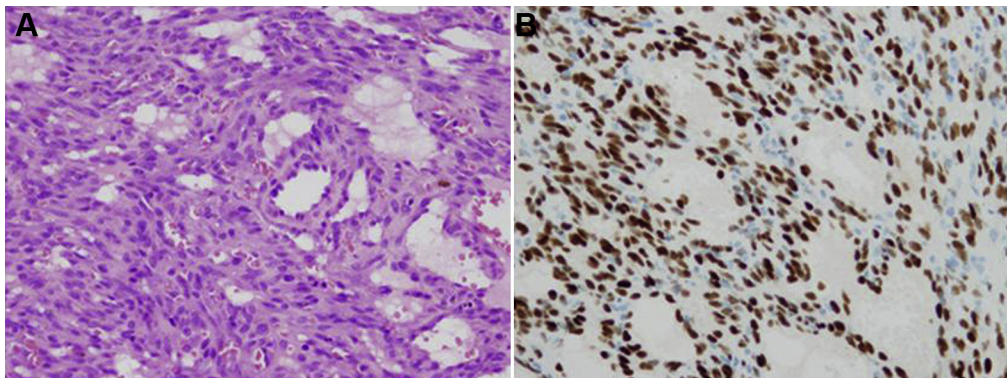
A debulking operation was planned based on a clinical diagnosis of recurrent hemangioma. Since preoperative blood tests revealed an elevated HbA1c level (7.8%) and the presence of syphilis, the



**Fig. 2.** MRI scan of the tongue lesion at the initial examination. A contrast-enhanced T1-weighted MRI scan showed only a small superficial lesion (arrows) on the dorsal surface of the tongue without infiltration into the deep tissue.

**Table 1**  
Laboratory data at the first examination.

Variable	Result
WBC ( $\times 10^3/\mu\text{L}$ )	3.2
Baso (%)	0.0
Eosin (%)	6.6
Neut (%)	64.0
Lymp (%)	14.7
Mono (%)	14.7
RBC ( $\times 10^6/\mu\text{L}$ )	3.74
Hgb (g/dL)	11.4
Hct (%)	33.6
PLT ( $\times 10^3/\mu\text{L}$ )	114
TP (g/dL)	7.5
Alb (g/dL)	3.9
Na (mEq/dL)	142
K (mEq/dL)	3.9
Cl (mEq/dL)	108
BUN (mg/dL)	15.3
Crea (mg/dL)	0.71
T-Bil (mg/dL)	0.3
AST (U/L)	12
ALT (U/L)	13
LD (U/L)	144
ALP (U/L)	231
CRP (mg/dL)	0.48
P-Glu (mg/dL)	101



**Fig. 3.** Histopathological findings of the biopsy specimen. (A) HE staining. A histopathological examination showed proliferation of monomorphic spindle cells with slit-like vascular containing erythrocytes. Original magnification  $\times 100$ . (B) Immunohistochemical staining for HHV-8. Positive reactions were seen in the tumor cells. Original magnification  $\times 100$ .

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