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(CASE REPORT)

Kaposi Sarcoma of the Tongue with Concomitant Cutaneous Involvement in an HIV-Negative Female Patient: A Case Report

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Abstract

Background*:* Kaposi sarcoma (KS) is an angioproliferative neoplasm associated with human herpesvirus 8 (HHV-8). It most commonly occurs in immunocompromised individuals, particularly those infected with human immunodeficiency virus (HIV). Oral involvement, especially of the tongue, is uncommon, and its occurrence in HIV-negative patients remains exceptionally rare.

Case Presentation: We report the case of a 54-year-old HIV-negative woman with a six-month history of progressive hyperpigmented cutaneous lesions involving the hands, feet, and thighs, associated with a violaceous angiomatous lesion affecting the anterior two-thirds of the tongue. Clinical examination also revealed cervical, axillary, and inguinal lymphadenopathy. Histopathological examination of a skin biopsy demonstrated a spindle-cell and vascular proliferation suggestive of Kaposi sarcoma. Immunohistochemical analysis confirmed the diagnosis with positive staining for HHV-8 and CD34. An inguinal lymph node biopsy further confirmed nodal involvement by Kaposi sarcoma. Contrast-enhanced computed tomography revealed multiple cervical, axillary, retroperitoneal, and inguinal lymphadenopathies. HIV serology was negative. The patient received weekly paclitaxel at a dose of 80 mg/m^2 for 12 cycles, achieving an excellent clinical response with significant regression of both lingual and cutaneous lesions and good treatment tolerance.

Conclusion*:* Lingual Kaposi sarcoma in HIV-negative patients is a rare clinical entity that may present with concomitant cutaneous and nodal involvement. Histopathological and immunohistochemical evaluation remains essential for diagnosis. Weekly paclitaxel represents an effective therapeutic option in advanced disease, providing favorable clinical outcomes. This case highlights the importance of considering Kaposi sarcoma in the differential diagnosis of vascular lesions of the tongue, even in the absence of HIV infection.

Keywords: Kaposi Sarcoma; Tongue; Oral Kaposi Sarcoma; HIV-Negative; HHV-8; Paclitaxel; Case Report

1. Introduction

Kaposi sarcoma (KS) is a vascular neoplasm of endothelial origin that is invariably associated with infection by human herpesvirus 8 (HHV-8), also known as Kaposi sarcoma-associated herpesvirus (KSHV) [1,2]. Since its first description by Moritz Kaposi in 1872, KS has been recognized as a heterogeneous disease encompassing four major clinical forms: classic, endemic (African), iatrogenic (transplant-associated), and epidemic (AIDS-related) KS [3,4]. The development of KS results from a complex interplay between HHV-8 infection, host immune status, genetic susceptibility, and inflammatory cytokine signaling pathways [5,6].

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Although the incidence of AIDS-related KS has declined considerably following the widespread introduction of highly active antiretroviral therapy (HAART), KS remains one of the most common vascular tumors worldwide [7,8]. The disease typically presents as multifocal violaceous macules, plaques, or nodules involving the skin and may affect mucosal surfaces, lymph nodes, and visceral organs [9,10]. Cutaneous lesions predominantly involve the lower extremities in classic KS, whereas disseminated mucocutaneous and visceral involvement is more frequently observed in immunocompromised patients [11].

Oral manifestations occur in approximately 20–70% of patients with AIDS-related KS and may occasionally represent the initial presentation of the disease [12,13]. The palate, gingiva, and oropharyngeal mucosa are the most frequently affected sites, whereas isolated tongue involvement remains exceedingly rare [14,15]. In HIV-negative individuals, oral KS is uncommon and may constitute a significant diagnostic challenge because of its resemblance to other vascular, inflammatory, or neoplastic lesions of the oral cavity [16,17].

The diagnosis of KS relies on histopathological examination demonstrating spindle-cell proliferation with irregular slit-like vascular spaces, extravasated erythrocytes, and hemosiderin deposits, supported by immunohistochemical detection of endothelial markers and HHV-8 latent nuclear antigen expression [18,19]. Treatment strategies depend on the extent of disease and may include local therapies, radiotherapy, systemic chemotherapy, immunomodulatory agents, or targeted approaches [20]. Paclitaxel has emerged as an effective treatment option for advanced or disseminated KS, producing durable responses in both HIV-positive and HIV-negative patients [21,22].

We report a rare case of lingual Kaposi sarcoma associated with cutaneous and nodal involvement in an HIV-negative woman. Given the exceptional nature of tongue involvement in the absence of HIV infection, this case highlights the importance of considering KS in the differential diagnosis of vascular lesions of the oral cavity and underscores the diagnostic value of histopathological and immunohistochemical evaluation.

2. Case Presentation

A 54-year-old woman with a history of type 2 diabetes mellitus presented with a six-month history of progressively enlarging hyperpigmented skin lesions involving the hands, feet, and thighs, associated with a painless violaceous lesion of the tongue. The symptoms evolved in the setting of unintentional weight loss without other constitutional complaints. Physical examination revealed an angiomatous violaceous lesion involving the anterior two-thirds of the tongue (Figure 1). Multiple hyperpigmented cutaneous lesions were observed on both hands, feet, and thighs. Palpation identified a left cervical lymph node measuring approximately 2 cm, bilateral axillary lymphadenopathy measuring 4 cm on the right and 3 cm on the left, as well as bilateral inguinal lymphadenopathy.

A skin biopsy was performed. Histopathological examination demonstrated a nodular spindle-cell and vascular proliferation suggestive of Kaposi sarcoma. Immunohistochemical analysis confirmed the diagnosis by demonstrating positive expression of HHV-8 and CD34 (Figures 3 and 4). To assess the extent of disease, an inguinal lymph node biopsy was subsequently performed and revealed a similar nodular spindle-cell vascular proliferation, confirming nodal involvement by Kaposi sarcoma. Contrast-enhanced cervicothoracoabdominopelvic computed tomography (CT) demonstrated multiple bilateral cervical lymph nodes measuring 7–15 mm, bilateral axillary lymphadenopathy measuring 8–15 mm, interaortocaval lymph nodes up to 10 mm, and bilateral inguinocrural lymph nodes ranging from 7 to 13 mm (Figure 2). No visceral organ involvement was identified. Serological testing for human immunodeficiency virus (HIV) was negative. Routine laboratory investigations and pre-treatment assessment were otherwise unremarkable. Given the disseminated mucocutaneous and nodal disease, systemic chemotherapy with weekly paclitaxel at a dose of 80 mg/m^2 was initiated. The patient received a total of 12 weekly administrations. Treatment was well tolerated, with no significant adverse events reported. Clinical evaluation during follow-up showed marked regression of the lingual lesion and cutaneous manifestations, accompanied by a substantial reduction in lymph node size, consistent with an excellent clinical response (Figure 1).

Figure 1 Serial clinical photographs showing the response of lingual Kaposi sarcoma to weekly paclitaxel chemotherapy. (a) Baseline lesion at diagnosis; (b) intermediate response; (c) marked regression after completion of treatment

2.1. Investigations

Laboratory investigations, including complete blood count, renal and liver function tests, and baseline pre-treatment assessment, were within normal limits. Serological testing for human immunodeficiency virus (HIV) was negative. To evaluate the extent of disease, an inguinal lymph node biopsy was performed and showed a similar spindle-cell vascular proliferation consistent with nodal involvement by Kaposi sarcoma.

Contrast-enhanced computed tomography (CT) of the neck, chest, abdomen, and pelvis revealed multiple bilateral cervical lymph nodes measuring 7–15 mm, bilateral axillary lymphadenopathy measuring 8–15 mm, interaortocaval lymph nodes up to 10 mm, and bilateral inguinocrural lymph nodes ranging from 7 to 13 mm (Figure 2). No evidence of visceral organ involvement was detected.

Histopathological examination of a skin biopsy revealed a nodular proliferation composed of spindle-shaped cells associated with irregular vascular channels, raising suspicion for Kaposi sarcoma. Immunohistochemical analysis demonstrated strong positivity for HHV-8 and CD34, confirming the diagnosis (Figures 3 and 4). Based on the clinical presentation, histopathological findings, immunohistochemical confirmation of HHV-8 infection, and radiological evidence of nodal dissemination, a diagnosis of disseminated Kaposi sarcoma involving the tongue, skin, and lymph nodes in an HIV-negative patient was established.

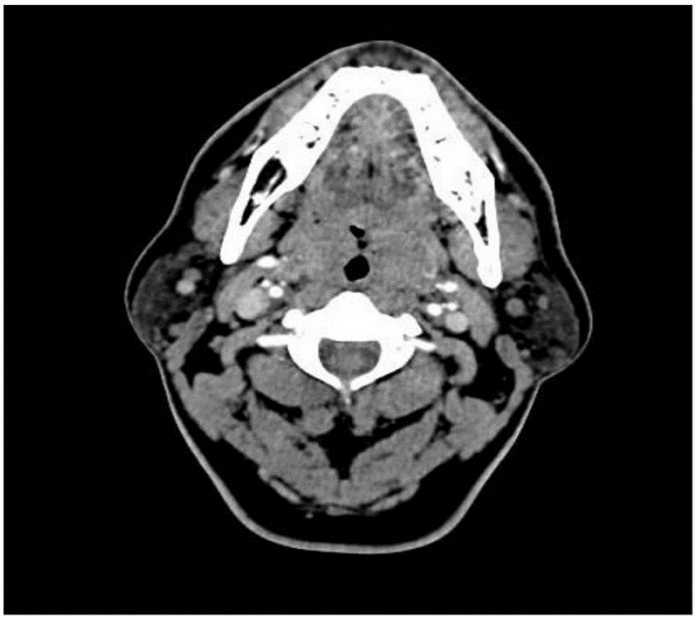

Figure 2 Contrast-enhanced axial CT scan of the neck showing diffuse enlargement and heterogeneous enhancement of the tongue associated with bilateral cervical lymphadenopathy.

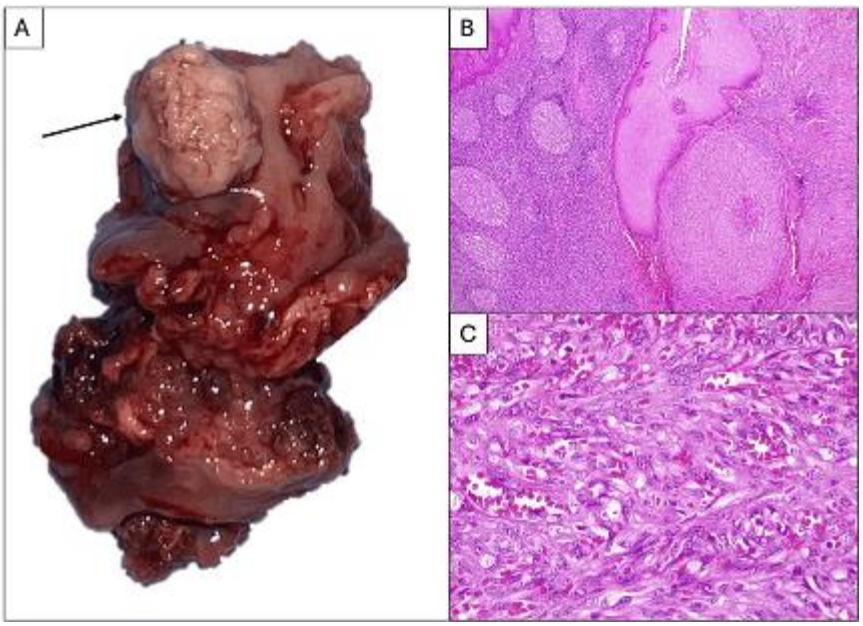

Figure 3 Gross pathological and histological features of Kaposi sarcoma. (A) Macroscopic appearance of the lymph node showing a nodular tumor lesion (arrow); (B) low-power photomicrograph demonstrating nodular spindle-cell proliferation; (C) high-power view showing spindle cells forming slit-like vascular spaces containing erythrocytes (H&E staining).

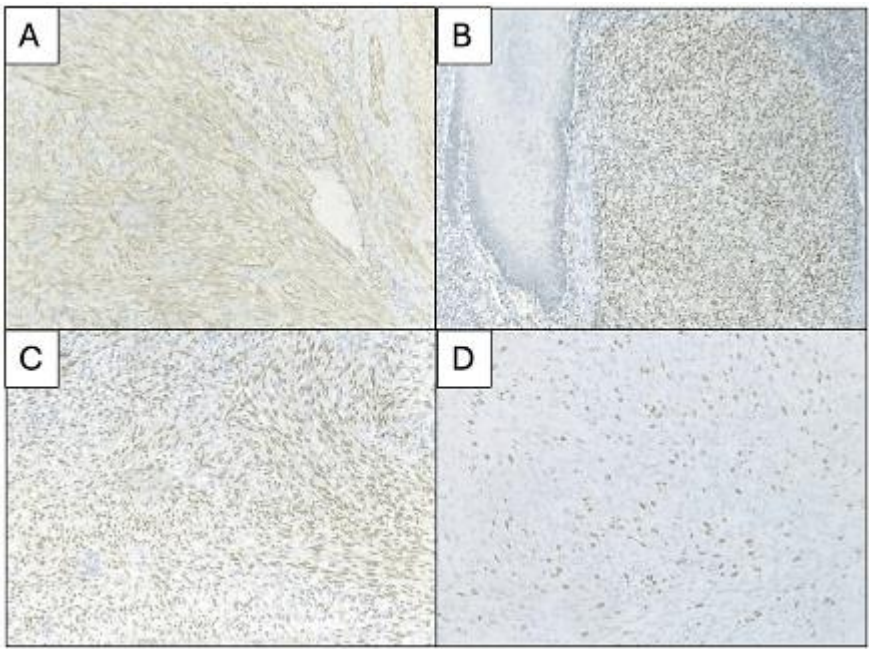

Figure 4 Immunohistochemical profile of Kaposi sarcoma. (A) CD34 expression in tumor cells; (B) nuclear positivity for HHV-8; (C) ERG nuclear expression confirming endothelial differentiation; (D) Ki-67 staining demonstrating the proliferative activity of the tumor.

2.2. Diagnosis

The diagnosis of disseminated Kaposi sarcoma involving the tongue, skin, and lymph nodes in an HIV-negative patient was established based on the combination of clinical, radiological, histopathological, and immunohistochemical findings.

Clinically, the patient presented with characteristic violaceous angiomatous lesions of the tongue associated with multiple hyperpigmented cutaneous lesions and generalized lymphadenopathy. Histopathological examination of the skin and lymph node biopsies revealed a spindle-cell vascular proliferation with features suggestive of Kaposi sarcoma. Immunohistochemical studies demonstrated positivity for HHV-8 and CD34, confirming the diagnosis.

Radiological assessment further documented cervical, axillary, retroperitoneal, and inguinal lymph node involvement without evidence of visceral dissemination. HIV serology was negative, supporting the diagnosis of non-HIV-associated (classic) Kaposi sarcoma with mucocutaneous and nodal involvement.

2.3. Treatment

Given the presence of disseminated disease involving the tongue, skin, and lymph nodes, the patient was considered a candidate for systemic therapy. After multidisciplinary evaluation, treatment with weekly paclitaxel at a dose of 80 mg/m^2 was initiated. Paclitaxel was administered intravenously once weekly, and the patient received a total of 12 cycles. The treatment was well tolerated, with no significant hematologic or non-hematologic toxicities observed during therapy. Clinical monitoring and routine laboratory assessments were performed throughout treatment.

A progressive improvement in the mucocutaneous lesions was noted during therapy, with marked regression of the lingual lesion and cutaneous manifestations. In addition, a significant reduction in the size of the palpable lymph nodes was observed. Following completion of treatment, the patient achieved an excellent clinical response with substantial resolution of disease manifestations (Figure 1).

2.4. Outcome and Follow-Up

The patient completed 12 weekly cycles of paclitaxel at a dose of 80 mg/m^2 without treatment interruption. Chemotherapy was well tolerated, and no significant hematologic or non-hematologic toxicities were observed during treatment. Clinical evaluation performed during and after completion of therapy demonstrated a marked regression of the lingual lesion, with substantial reduction in its size and vascular appearance. A significant improvement in the cutaneous lesions was also observed, characterized by flattening and fading of the hyperpigmented lesions. In addition,

a noticeable decrease in the size of the cervical, axillary, and inguinal lymph nodes was documented on physical examination.

The overall response was considered an excellent clinical response, with near-complete resolution of the oral lesion and marked improvement of the cutaneous manifestations (Figure 1). The patient's general condition improved, with stabilization of body weight and resolution of disease-related symptoms. At the last follow-up visit, the patient remained clinically stable with no evidence of disease progression or treatment-related complications. Continued clinical surveillance was recommended, including regular physical examinations and radiological assessment when clinically indicated, to monitor for potential recurrence or disease progression.

3. Discussion

Kaposi sarcoma (KS) is a vascular neoplasm associated with human herpesvirus 8 (HHV-8) infection and is classically categorized into four clinical variants: classic, endemic, iatrogenic, and epidemic (AIDS-related) KS [1–4]. Although KS is strongly associated with immunosuppression, particularly HIV infection, cases occurring in HIV-negative individuals continue to be reported and may pose significant diagnostic challenges [5,6]. The present case is remarkable because of the uncommon involvement of the tongue in an HIV-negative woman. Oral manifestations are frequently encountered in AIDS-related KS, with the palate and gingiva representing the most commonly affected sites [12,13]. In contrast, tongue involvement is rare, especially in HIV-negative patients [14,15]. Recent reports by Andrade-Carmona et al. and Mouna et al. highlighted the exceptional nature of lingual KS in HIV-negative individuals and emphasized the importance of considering this diagnosis when evaluating vascular lesions of the oral cavity [16,17]. Our case further contributes to the limited literature by documenting lingual KS associated with both cutaneous and nodal involvement in an HIV-negative female patient.

The diagnosis of oral KS may be challenging because its clinical appearance overlaps with several benign and malignant conditions, including hemangioma, pyogenic granuloma, bacillary angiomatosis, angiosarcoma, and lymphoma [15,16]. In the present case, the diagnosis was established through histopathological examination and immunohistochemical analysis. Histologically, KS is characterized by a proliferation of spindle-shaped cells forming irregular slit-like vascular spaces associated with erythrocyte extravasation [18]. Immunohistochemical positivity for HHV-8 remains the diagnostic hallmark and allows differentiation from other vascular neoplasms [18,19].

Although classic KS generally follows an indolent course and predominantly affects the lower extremities of elderly individuals of Mediterranean or Eastern European descent [3,11], disseminated mucocutaneous and nodal involvement may occur. Lymph node involvement, as observed in our patient, is less common in classic KS and is often associated with a more extensive disease burden requiring systemic treatment [9,11]. Computed tomography played an important role in assessing disease extent and confirming the presence of multiple lymph node stations without visceral organ involvement. The management of KS depends on disease extent, symptoms, cosmetic impairment, and the patient's immune status [20]. Localized lesions may be treated with surgery, radiotherapy, cryotherapy, or intralesional therapies, whereas disseminated disease generally requires systemic treatment [20]. Taxanes have demonstrated significant activity in advanced KS. In a multicenter phase II trial, paclitaxel achieved high response rates and durable disease control in patients with advanced KS [21]. Subsequent studies confirmed the efficacy and favorable safety profile of paclitaxel in both HIV-positive and HIV-negative patients [22]. Given the multifocal mucocutaneous lesions and nodal dissemination observed in our patient, systemic chemotherapy with weekly paclitaxel was selected, resulting in excellent clinical response and good tolerance.

This case highlights several important clinical messages. First, Kaposi sarcoma should be considered in the differential diagnosis of persistent vascular lesions of the tongue, even in HIV-negative individuals. Second, histopathological and immunohistochemical evaluation, particularly HHV-8 staining, remains essential for establishing the diagnosis. Finally, weekly paclitaxel represents an effective therapeutic option for disseminated KS and may achieve substantial clinical benefit with acceptable toxicity.

In conclusion, lingual Kaposi sarcoma associated with cutaneous and nodal involvement in an HIV-negative woman is an exceptionally rare presentation. Reporting such cases contributes to improving recognition of atypical manifestations of KS and supports the role of systemic chemotherapy in advanced disease.

4. Conclusion

Kaposi sarcoma involving the tongue is an exceptionally rare manifestation, particularly in HIV-negative patients. We report a rare case of lingual Kaposi sarcoma associated with cutaneous and nodal involvement in a 54-year-old HIV-negative woman. The diagnosis was established through histopathological examination and immunohistochemical confirmation of HHV-8 expression.

This case highlights the importance of considering Kaposi sarcoma in the differential diagnosis of persistent vascular lesions of the oral cavity, even in the absence of HIV infection or other evident causes of immunosuppression. Comprehensive clinical evaluation, imaging studies, and immunohistochemical analysis are essential for accurate diagnosis and staging. Systemic treatment with weekly paclitaxel resulted in an excellent clinical response and was well tolerated, supporting its role as an effective therapeutic option for disseminated disease. Reporting such uncommon presentations contributes to increasing awareness of atypical forms of Kaposi sarcoma and may facilitate earlier diagnosis and appropriate management.

Compliance with ethical standards

Disclosure of conflict of interest

No conflict of interest to be disclosed.

Statement of informed consent

Informed consent was obtained from all individual participants included in the study.

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