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A Case of Ocular Kaposi's Sarcoma Successfully Treated with Highly Active Antiretroviral Therapy (HAART) Combined with Docetaxel

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Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
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Conflict of interest: None declared

Patient: Male, 24
Final Diagnosis: Ocular Kaposi's sarcoma
Symptoms: Eyelid swelling • red eye
Medication: —
Clinical Procedure: Biopsy
Specialty: Oncology

Objective: Rare disease

Background: Ocular Kaposi's sarcoma (KS) involving the conjunctiva and ocular adnexa is uncommon and is usually treated with cryotherapy or surgical excision. We report a case of ocular KS successfully treated with HAART combined with 8 cycles of weekly docetaxel.

Case Report: Our patient was a 24-year-old, treatment-naïve, HIV-positive (CD4 cell count 198 cells/mm^3), homosexual man treated as having atypical hordeolum and subconjunctival hemorrhage, and later confirmed with pathology to have ocular KS with immunohistochemistry study showing KS with positive HHV8, CD34, CD31, and focal positive staining with Factor VIIIRA. HAART therapy was initiated combined with weekly docetaxel. With 2-month treatment of HAART and 8 cycles of weekly docetaxel, the KS of the bulbar conjunctiva and the eyelid partially resolved.

Conclusions: HAART combined with weekly docetaxel is an effective and well-tolerated option for ocular KS, which could be considered before cryotherapy or surgical excision.

MeSH Keywords: Anti-Retroviral Agents • Antineoplastic Agents • Herpesvirus 8, Human • Sarcoma, Kaposi

Full-text PDF: https://www.amjcaserep.com/abstract/index/idArt/910374

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Background

Kaposi's sarcoma (KS) is the most frequent malignancy associated with human immunodeficiency virus (HIV) infection. It is categorized into 4 different types: classic, endemic, epidemic, AIDS-associated, and immunosuppression therapy-related. Epidemic KS was first described in 1872 and KS-associated herpes virus was first detected in 1994 [1,2]. It frequently involves mucocutaneous sites, such as the lower extremities, face, trunk, genitalia, and oropharynx. KS also commonly involves lymph nodes and visceral organs, most notably the respiratory and gastrointestinal tracts. However, involvement of the conjunctiva and ocular adnexa are uncommon manifestations of KS [3].

External ocular KS may present as a mass lesion on the eyelid, which is easily misdiagnosed as a hordeolum, chalazion, or simply as a subconjunctival hemorrhage (a hordeolum usually is caused by infection, whereas a chalazion is caused by non-infectious meibomian gland occlusion). Cryotherapy and surgical excision are mainstay treatments for eyelid KS and bulbar conjunctiva KS, respectively [4].

Case Report

A 24-year-old, treatment-naïve, HIV-positive, homosexual man came to our attention for bilateral red eyes with eyelids swelling. An ophthalmologic exam revealed atypical hordeolum and subconjunctival hemorrhage. Bilateral intraocular pressures were 9 mmHg with visual acuity 20/30. Two months later, the patient was admitted to the hospital for productive cough associated with shortness of breath, subjective fever, loss of appetite, and weight loss. The subconjunctival hemorrhage was attributed to thrombocytopenia, with the lowest platelet count dropping below 20 000. A CT scan of the chest revealed bilateral, diffuse, patchy infiltrates (Figure 1). Bronchoscopy was attempted but aborted because of an inaccessible pharynx due to profound edema friability of the surrounding tissue. At presentation, viral load was 157 554 copies/ml and the CD4 cell count was 198 cells/mm^3.

On physical exam, a 2.5×2×1 cm vascular, pedunculated, soft, friable mass on anterior maxillary gingiva between teeth 6 and 8 was noticed and biopsied by oral and maxillofacial surgery (Figure 2). The pathology with immunohistochemistry study showed KS with positive HHV8, CD34, and CD31 and focal positive staining with Factor VIIIRA. AIDS-related KS was staged T1I1S1 [5]. HAART therapy (elvitegravir/cobicistat/emtricitabine/Tenofovir alafenamide) was initiated with docetaxel. One month later, the viral load decreased to 412 copies/ml and CD4 cell count increased to 218 cells/mm^3. With 2 months treatment of HAART and 8 cycles of weekly docetaxel, KS of the bulbar conjunctiva and the eyelid partially resolved (Figure 3). Four months later, viral load decreased to 98 copies/ml and CD4 cell count increased to 299 cells/mm^3. This patient responded very well to the regimen of HAART combined with docetaxel, which avoided surgical treatment and cryotherapy for the bulbar conjunctiva and eyelids KS.

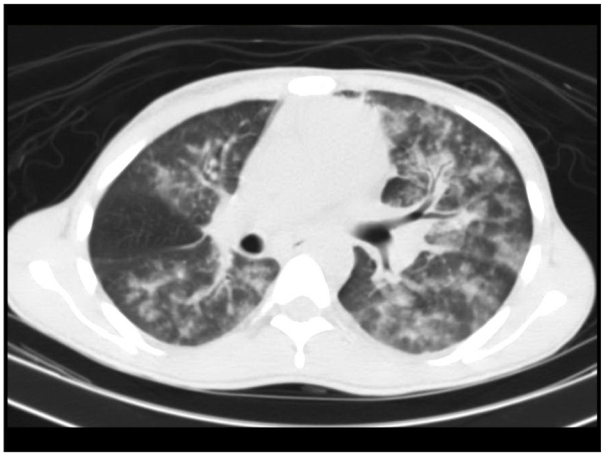

Figure 1. Chest CT scan revealed bilateral diffuse patchy infiltrates due to Kaposi's sarcoma.

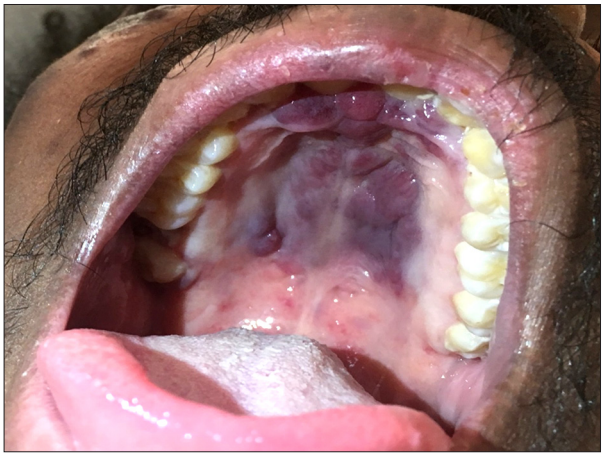

Figure 2. Oral involvement of Kaposi's sarcoma (post-biopsy of a pedunculated gingival mass located at anterior maxillary gingiva between teeth 6–8).

Discussion

With widespread use of HAART in the United States, the incidence of KS has remarkably declined and ocular KS has become a rare manifestation of the disease [6–9]. We described our experience of favorable clinical response in a patient who was treated with HAART and docetaxel with no serious adverse effects. This treatment resulted in complete resolution of the oral KS of the palate and gingiva, as well of respiratory symptoms.

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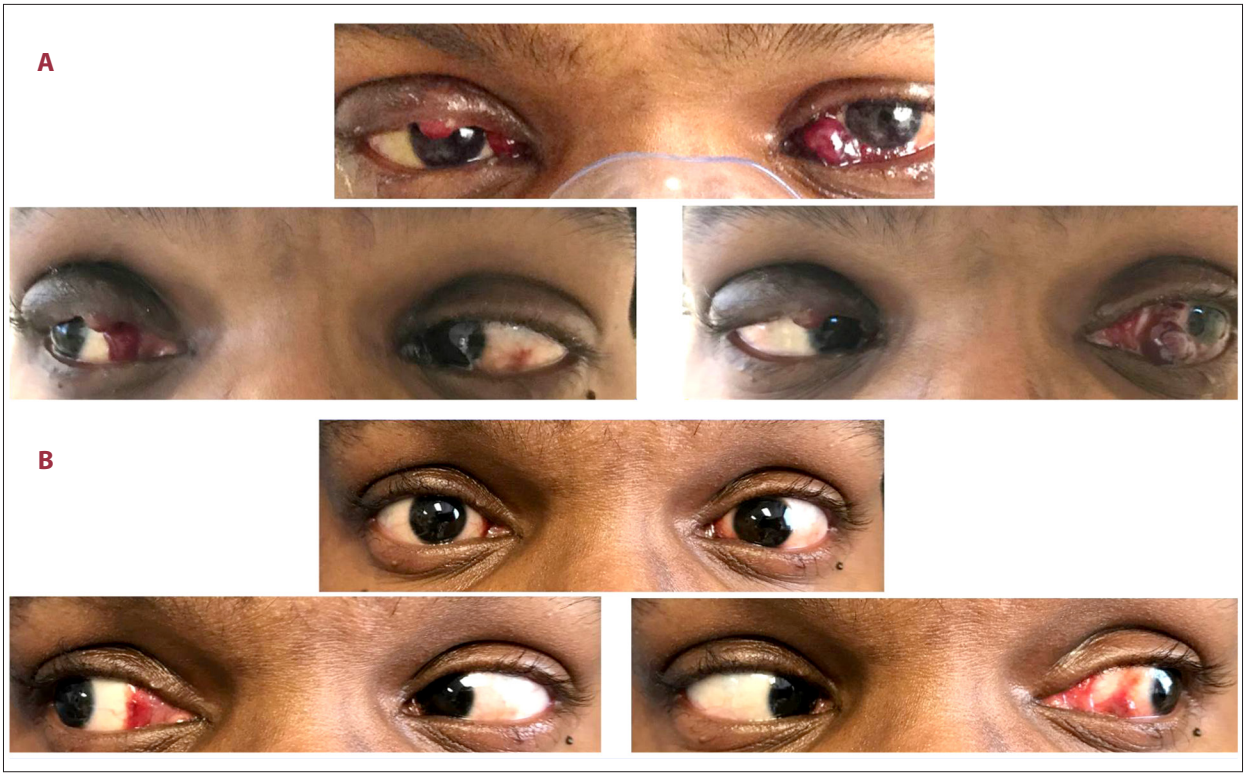

Figure 3. Comparison between pretreatment and post-treatment of Kaposi's sarcoma (KS). (**A**) KS on the right swollen eyelid with bilateral bulbar conjunctival KS. (**B**) Eyelid KS and bulbar conjunctival KS responded very well to 2-month treatment with HAART and 8 cycles of weekly docetaxel.

KS-associated herpes virus (KSHV, also known as HHV8) has been proposed to be the causative agent for this neoplasm. However, HHV-8 in conjunctival or eyelid involvement of KS has been rarely reported [10,11]. The biopsy of this KS case exhibited expressivity of HHV8, as well as CD 31, CD34, and Factor VIII-related antigen. Release of growth factors and pro-inflammatory cytokines like interlukin-6, which regulates oncogene expression through STAT3-dependent mechanism, is proposed to be dysregulated along with compromised immune surveillance in HIV-infected hosts [12,13]. In addition, cross-activation of oncogenic HHV8 via HIV-tat transcription factors further results in dysregulated expression of oncogenes and tumor suppressor genes, leading to angiogenic properties of the cell [14]. Thus, treatment target for such individuals involves reducing HIV viral load to subsequently decrease tumor burden, and also directly targeting the cancer cells.

Docetaxel inhibits microtubular depolymerization, which stabilizes microtubules and arrests the cell in G2M phase of the cell cycle. In addition, docetaxel attenuates the effects of Bcl-2 and Bcl-xL gene expression, which promotes a cascade of events that ultimately leads to apoptotic cell death [15]. This anti-neoplastic property of docetaxel combined with anti-retroviral therapy showed promising results for KS-associated ocular lesions. Docetaxel is commonly used to treat breast cancer, head and neck cancer, stomach cancer, prostate cancer, and non-small-cell lung cancer. Our report supports the findings of a previous study that concluded weekly docetaxel is safe and effective in the treatment of advanced-stage AIDS-related KS [16]. HAART has been associated with objective tumor response in patients with AIDS-related KS, presumably by augmenting the immune status of the host, with subsequent immune control of the underlying HHV8 infection and KS. Our experience suggests that HAART combined with weekly docetaxel is an effective and well-tolerated option for treating ocular KS.

Conclusions

With the advent of HAART, KS-associated with HIV-1 infection as the initial clinical manifestations has become rare. HAART combined with weekly docetaxel is an effective and well-tolerated option for treatment of ocular KS, which should be considered before cryotherapy or surgical excision.

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