

A Rare Recurrent Kaposi Sarcoma in an HIV-Seronegative Saudi Man: A Case Report and Literature Review

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Abstract

Introduction: Kaposi sarcoma is a well-recognized vascular lesion that primarily affects the skin, following the course of three stages; patch, plaque and nodules. It is strongly associated with human herpes virus 8 (HHV8) and Human immunodeficiency virus (HIV). Nevertheless, it is a rare presentation in immunocompetent individuals. **Case presentation:** A seronegative middle-aged Saudi man with history of nasal Kaposi sarcoma, presents with a recurrent dusky brown nodular lesion on the thigh. Histopathologic examination showed a spindle cell lesion with intervening slit-like vascular spaces that are immunoreactive to HHV8. **Conclusion:** Kaposi sarcoma can occur and recur in the setting of HIV-seronegative healthy individuals, whom they present with a nodular vascular skin lesion. We recommend the assessment of HHV8 status in such lesions, despite seronegative HIV status.

Keywords: Kaposi sarcoma, human herpesvirus 8.

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INTRODUCTION

In the late eighteen-hundreds, Dr. Moritz Kaposi was the first to ever describe Kaposi sarcoma, with the title “*idiopathic multiple pigmented sarcoma of the skin*”, he described it in his paper as follows; “*nodules the size of shot, peas or hazelnuts and brown-red to blue-red in color, develop in the skin without a known general or local cause*” [1]. Decades gone by, now Kaposi sarcoma has been better understood in terms of its clinical course and pathogenesis. The Herpes-human virus 8 (HHV8) has been detected in all the clinical forms of Kaposi sarcoma [2]. It can occur in any site, and its progression occurs through three stages:

Macular, plaque and ultimately *tumor* stage. It has been further categorized into four distinct clinic epidemiological settings that range from indolent to highly aggressive:

- i. Classic Kaposi sarcoma: in elderly men of Mediterranean and Eastern European descent.
- ii. Endemic African Kaposi sarcoma
- iii. Kaposi sarcoma in immunosuppressed patients
- iv. HIV and AIDs-related Kaposi sarcoma [2, 3].

CASE PRESENTATION

In 2008, a 35-year-old Saudi man has been diagnosed with Kaposi sarcoma of the nasal mucosa, following a nasal excisional biopsy in a private hospital (*Figure 1*). Fourteen years later, he presents to King Fahad Specialist hospital in Dammam, Saudi Arabia, complaining of multiple nodular lesions at the medial aspect of his right thigh, associated with mild bleeding after trauma, that he noticed three months back. On physical examination, the right thigh showed dusky-red skin nodules with crusted yellow scales on the surface, ranging in size from 0.5 cm to 1.5 cm. HIV-1, HIV-2 and hepatitis seroprofiles were all negative. PETCT scan revealed skin nodules in the antero-medial aspect of the distal right thigh in keeping with a known history of Kaposi sarcoma, with no evidence of distant or nodal metastasis.

A skin punch biopsy of the right thigh lesion has been performed (*Figure 2*). Subsequently, a wide local excision of the right thigh lesion followed. The patient is currently under routine follow-up with radiation oncology clinic every six months to monitor for recurrence and evaluate the potential need for radiotherapy.

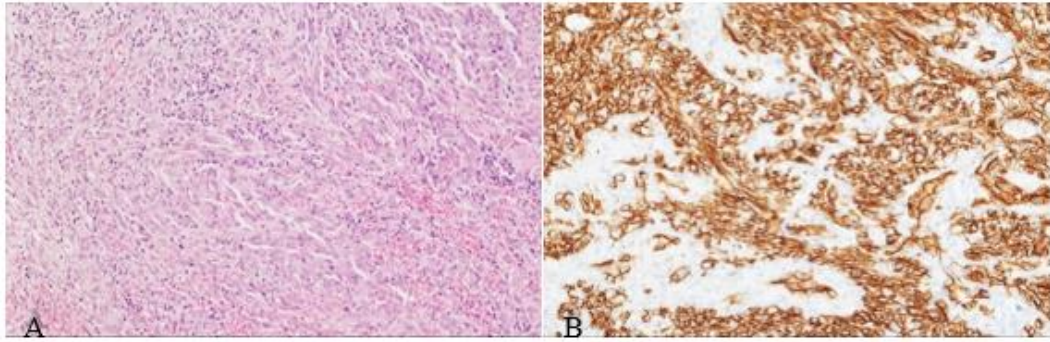


Figure 1: (A) H&E, intermediate power view of a spindle cell proliferation arranged in intersecting fascicles with intervening slit-like vascular spaces and extravasated erythrocytes. (B) CD34 immunostain, strong diffuse positivity

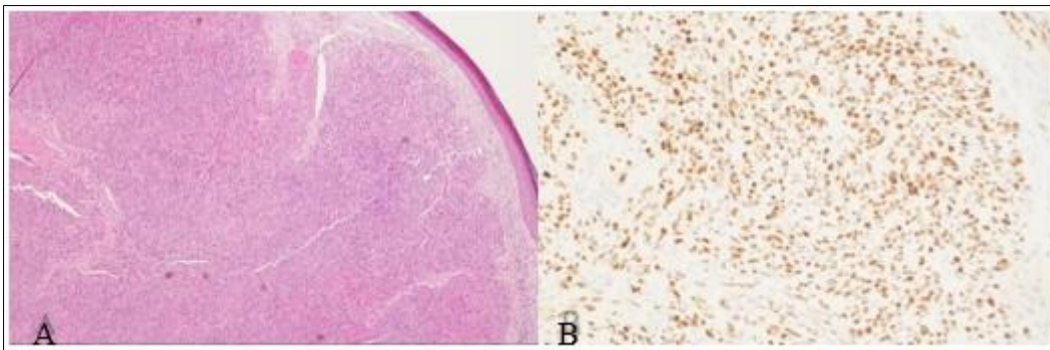


Figure 2: (A) H&E, low power view of a dermal based spindle cell lesion composed of spindle cells arranged in intersecting fascicles with intervening slit-like vascular spaces. (B) HHV8 immunostain, positive with a stippled nuclear pattern

Macroscopically, a dusky-brown fungating mass with surface ulceration found at the skin surface (Figure 3), measured 2.3 cm in maximum dimension,

along with two smaller grey nodules surrounding the index mass, measuring 0.3 and 0.8 cm each, respectively. All the surgical resection margins were negative.

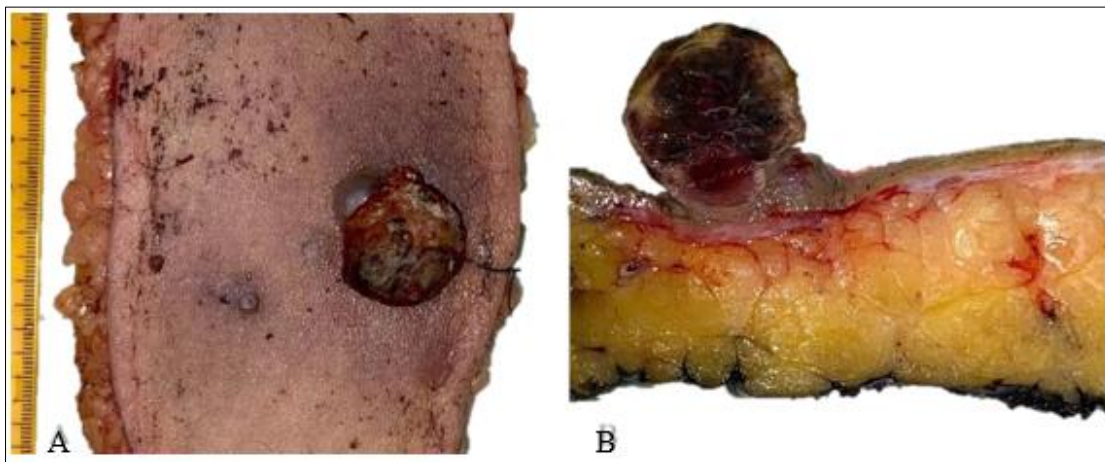


Figure 3: (A) Gross photograph of a dusky brown fungating mass with an ulcerated surface, surrounded by grey-colored smaller nodules. (B) Cross-section of the lesion with a nodular heterogenous cut surface filled with blood

Microscopically, a well-circumscribed nodular lesion with surface ulceration, composed of uniform spindle cells arranged in intersecting fascicles with intervening variably sized slit-like vascular channels, frank mitoses and extraverted red blood cells and

hemosiderin were also noted in a background of inflammation and necrosis (figure 4). Well-controlled HHV-8 immunostain was performed, revealing a strong diffuse and stippled nuclear pattern of staining (figure 4).

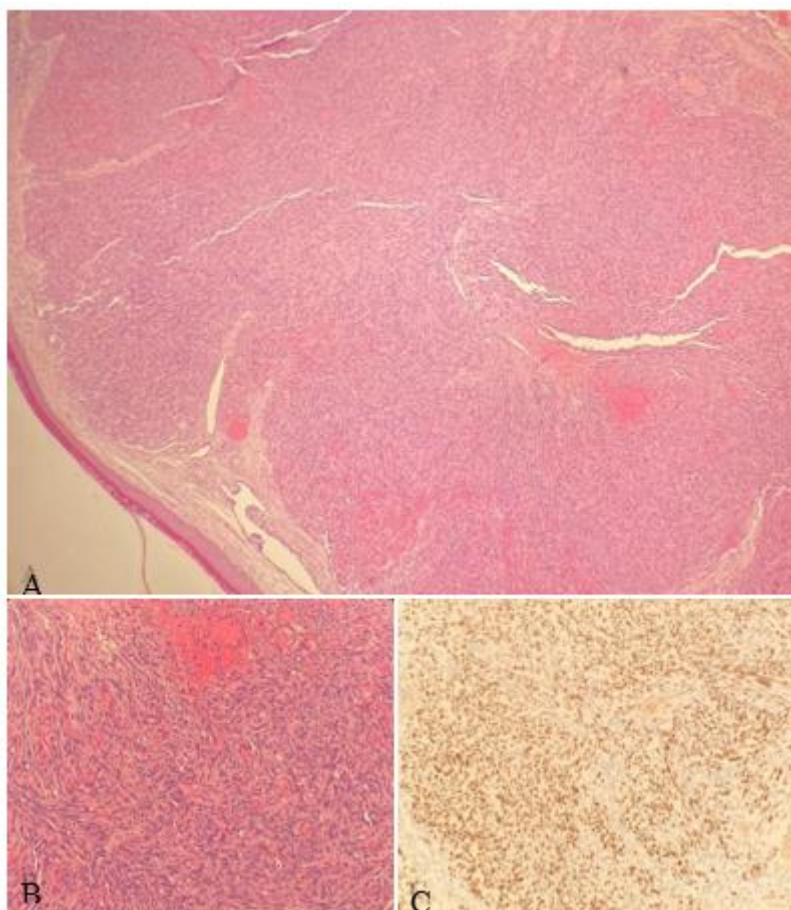


Figure 4: (A and B) H&E, low and high power view of a spindle cells with intervening slit-like vascular channels surrounded by lymphoplasmacytic infiltration and occasional mitotic figures. (C) HHV8 immunostain, demonstrates strong nuclear positivity to HHV8

DISCUSSION

This case depicts the recurrence of KS in an HIV-seronegative and immunocompetent patient. The prevalence of Kaposi sarcoma in the Arab countries is underreported, particularly the recurrence rates of KS; the utter lack of documented cases in the region has prompted the authors to publish this report. In view of the reported cases in other regions of the world, a recurrent classic KS in a 69-year-old Japanese man has been reported by Kusakari *et al.*, diagnosed based on morphology, immunoreactivity to HHV8 and PCR-based detection of HHV8 DNA [4]. Another recurrent case of classic Kaposi sarcoma has been reported in Iran by Etesami *et al.*, occurring in a 43-year-old man with multiple ear nodules, which displayed immunoreactivity to HHV8 too [5]. Etesami *et al.*, reviewed 24 different cases of Kaposi sarcoma of the ear and concluded their article by establishing the ear as a predilection site for head and neck HIV-seronegative patients [5]. In the contrary to our case, the patient was diagnosed with KS of the nasal mucosa, followed by KS of the cutaneous thigh region.

Furthermore, another paper has reported the first pediatric Kaposi sarcoma case in Iraq, in a six-year-

old, HIV-seronegative boy, he was diagnosed with rapidly progressive Kaposi sarcoma on the basis of skin biopsy findings. According to Al-kzayer *et al.*, the nodules developed after long-term use of valproic acid, in which they have concluded their article by documenting the causal relationship between valproic acid therapy and developing Kaposi sarcoma [6].

Two studies were conducted in Saudi Arabia to assess the prevalence of HHV8. In one hospital-based study by AlMuneef *et al.*, HHV8 was assessed in the general Saudi population on both renal failure patients and in patients without renal disease, the rates were 7% and 4%, respectively [7]. The second study by Alzahrani *et al.*, confirmed high HHV8 rates (18%) in renal failure patients, relative to its prevalence in the general population (1.7%). Interestingly, none of the HHV8 seropositive patients in their study, developed Kaposi sarcoma [8].

CONCLUSION

Given the scarcity of studies and the absence of reported recurrent Kaposi sarcoma cases in the Arab region, in the setting of HIV-seronegative patients, this case report aims to contribute to the data and help further

identify such lesions and prepare clinicians and pathologists to the possibility of encountering such entity in otherwise healthy individuals. In conclusion, it's plausible to keep Kaposi sarcoma in the differential diagnoses of all vascular skin lesions in immunocompetent individuals and the importance of assessing seropositivity of all vascular lesions toward HHV8 in the proper clinical and histopathologic settings, despite the patient's seronegative HIV status.

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