



Case Report

Osseous Metastases in HIV Patient with Kaposi Sarcoma

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Abstract

Osseous lesions in AIDS-related Kaposi sarcoma (KS) are extremely rare, with approximately 30 published cases to date. We highlight a case of a HIV-positive patient who was initially misdiagnosed with cellulitis but later found to have KS with osseous metastases and concomitant osteomyelitis. MRI was necessary to detail unique features and confirm both pathologies. However, biopsy of a lesion is still the gold standard for definitive diagnosis of KS and can be confirmed with immunohistochemistry for LANA1. The mainstay of treatment is a tailored combination of ART, chemotherapy, local radiation therapy, and surgery. Given that osseous KS is rare and often confounded with other musculoskeletal pathology, education is critical for proper and timely treatment. Our case report provides trainees and physicians with clinical, radiographic, and histopathologic images to aid in the diagnosis and management of osseous KS.

Keywords: Kaposi Sarcoma; Osseous Metastases; HIV; HHV8

Introduction

Kaposi sarcoma (KS) is an angioproliferative neoplasm commonly associated with human herpesvirus 8 (HHV8) and HIV/AIDS. It often presents as violaceous pink to purple plaques on the skin or mucocutaneous surfaces with localized lymphedema. Osseous lesions in AIDS-related KS are extremely rare, with approximately 30 published cases to date [1]. Patients often present with bone pain and have osseous KS lesions in the axial and/or maxillofacial skeleton [1]. We highlight a case of a HIV-positive patient who was found to have KS with osseous metastases and concomitant osteomyelitis.

Case Report

A 40-year-old Black man with HIV presented with three months of left lower extremity swelling, pain, and worsening rash. He had been inconsistently taking his antiretroviral therapy (ART) due to financial difficulties and was treated at an outside hospital around 3 weeks prior for suspected cellulitis. He reported

improvement in the leg swelling during that hospitalization, but the swelling recurred upon discharge due to inability to afford his antibiotics. He also reported a history of intermittent melena and rectal bleeding. No fevers, night sweats, or unintentional weight loss.

On admission, he was afebrile with a normal white blood cell count (WBC), CD4 201 (12.5%), and viral load 671 copies/mL. His exam was positive for small 1-2 mm red-purple papules on his hard palate (Figure 1a), axillary and inguinal lymphadenopathy, violaceous nodules and plaques with substantial lymphedema of his left thigh (Figure 1b), and blistering of his left third toe with tenderness to palpation at the base. Initial CT of his left lower extremity demonstrated soft tissue swelling and erosive osseous changes in his second and third toes as well as multiple new scattered lytic lesions in the lower spine and pelvis. Subsequent MRI of his lumbar spine (Figure 2a) and pelvis (Figure 2b) re-demonstrated numerous enhancing osseous lesions throughout his lumbar spine, partially visualized thoracic spine, and pelvis. MRI of his left foot also confirmed acute osteomyelitis of his second through fifth distal and middle phalanges.



Figure 1: Dermatological manifestations of Kaposi sarcoma. (a) Red-purple small papules on hard palate. (b) Violaceous nodules and plaques with substantial lymphedema and peau d'orange changes.



Figure 2: MRI lumbar spine and pelvis. (a) Multiple enhancing lesions throughout the axial and appendicular skeleton with the largest lesions within the left aspect of S1 measuring up to 1.5 cm; and within the left aspect of the L1 vertebral body extending into the posterior elements measuring up to 1.9 cm. (b) Multifocal PD FS hyperintense and T1 iso-hypointense enhancing intramedullary lesions throughout the pelvis.

Shave biopsy with hematoxylin and eosin staining was performed on a left thigh lesion, which revealed monomorphic spindle cells with slit-like spaces, positive promontory sign (Figure 3a), and diffusely positive HHV8 immunohistochemical stain consistent with KS (Figure 3b). The workup for primary hematological malignancy was negative, and infectious disease workup was also unremarkable. Complete imaging with CT chest, abdomen, and pelvis was negative for metastases to the gastrointestinal (GI) tract, lungs, liver, and spleen.

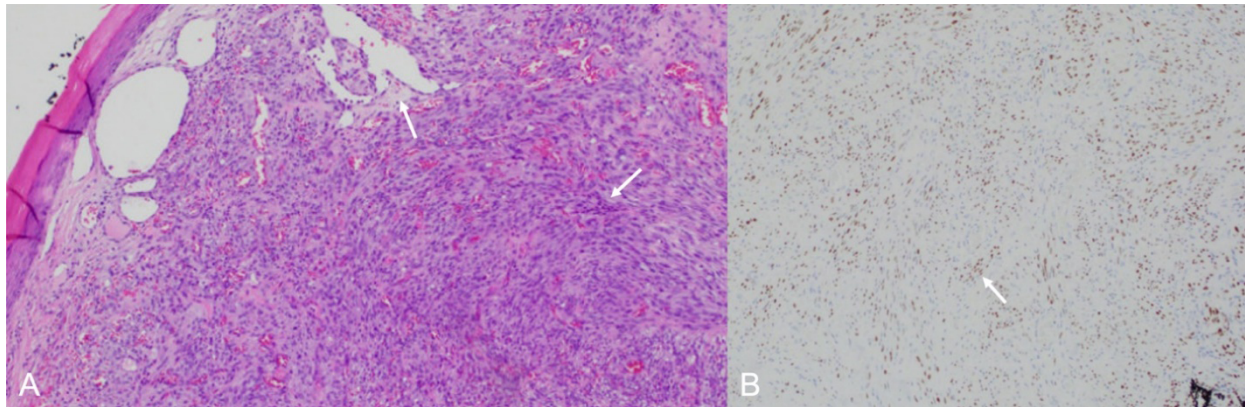


Figure 3: Left thigh biopsy results. (a) Hematoxylin and eosin staining showing monomorphic spindle cells with slit-like spaces and promontory sign. (b) HHV8 immunohistochemical stain highlighting the neoplastic spindle cells. Findings are consistent with suspected clinical diagnosis of Kaposi sarcoma.

Patient was scheduled for outpatient chemotherapy and ART follow-up. He was also advised to obtain an esophagogastroduodenoscopy and colonoscopy to evaluate for visceral KS. He remained stable throughout hospitalization and was enrolled in a patient assistance program for his antiretroviral therapy and antibiotics for his osteomyelitis.

Discussion

AIDS-related KS is the second most common tumor in HIV patients with CD4 counts less than 200 cells/mm³ after non-Hodgkin's lymphoma [2, 4]. In these patients, KS tends to be more aggressive and widespread, often involving the skin, oral mucosa, lymph nodes, GI tract, lungs, liver, and spleen [5]. Osseous involvement of AIDS-related KS is rare, and the mechanisms underlying the development of these lesions remain poorly understood. However, when osseous metastases do occur, they most often affect the axial skeleton [1,5]. In a systemic review by Caponetti et al., he noted that AIDS-related KS with lesions in the pelvis also all had concomitant involvement of the spine. In addition, the level of bone pain reported by patients appeared to be unrelated to the extent of the bone lesion and did not usually represent direct invasion from an adjacent cutaneous lesion [1]. These findings were consistent with the presentation of our patient.

The diagnosis of osseous metastases in KS can be challenging due to its rarity and the similarity of radiological findings to other bone metastases. In settings where KS is less prevalent, a high index of suspicion is crucial for timely diagnosis. Radiographically, most AIDS-related KS bone lesions appear osteolytic [1]. The preferred imaging modality to identify lytic bone changes is CT [6,7] and the differential diagnosis often

includes bacillary angiomatosis, angiosarcoma, osteomyelitis, mycobacterial infections, hemangioendothelioma, and primary bone lymphoma [2,3]. MRI can better differentiate KS from these etiologies by highlighting its unique features – discontinuous, yet well-defined, multifocal or polyostotic osteolysis, enhancing soft tissue masses, and the absence of joint involvement [3]. Our patient was unique because he had both osseous KS lesions and osteomyelitis, so MRI was necessary to detail both pathologies. To our knowledge, our case is the only report to date of concurrent osseous KS and osteomyelitis. Biopsy of a lesion is still the gold standard for definitive diagnosis of KS.

Histologically, KS is characterized by spindle cell proliferation of irregular, complex vascular channels in the form of slit-like spaces (positive promontory sign) with extravasated red blood cells and hyaline globulins. The presence of HHV8 can be confirmed with immunohistochemistry for LANA1. Silver staining with Warthin Starry Grocott can also differentiate between bacillary angiomatosis (positive) and KS (negative) [2,3].

The management of osseous metastases in KS requires a multidisciplinary approach, and the primary goals include pain management, preservation of skeletal integrity, and a tailored combination of ART, chemotherapy, local radiation therapy, and surgery [1,3,8]. The prognosis of osseous metastases in KS is generally poor, reflecting the advanced stage of disease at the time of diagnosis [1,8]. Despite aggressive treatment approaches, the overall survival is limited, [9] and the focus often shifts towards improving quality of life and managing symptoms. Regular follow-up is essential to monitor disease progression, manage treatment-related side effects, and address emerging complications.

Conclusion

Our case report provides trainees and physicians with clinical, radiographic, and histopathologic images to aid in the diagnosis and management of osseous KS. Our patient was initially misdiagnosed with cellulitis, so further workup was not obtained until subsequent presentation. Given that osseous KS is rare and often confounded with other musculoskeletal pathology, education on cutaneous lesion presentation, radiographic features, and dermatology pathology is critical for proper and timely treatment.

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Authors Contributions: All authors had full access to the data presented and a role in writing the manuscript.

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