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Case report

Kaposi sarcoma with musculoskeletal manifestations in a well-controlled HIV patient

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ABSTRACT

We present here a case of relapsed HIV-related Kaposi Sarcoma (KS), manifesting as a plantar ulcer with underlying bone involvement in a patient with well-controlled HIV. Radiographic and magnetic resonance imaging of the patient's right foot showed bone destruction suggestive of osteomyelitis. However, when a bone biopsy was done, this was consistent with KS, without any signs of bone infection. Patient was initially diagnosed with KS four years prior. He was successfully treated at the time with doxorubicin, radiation therapy, and began HIV therapy. At the time of the KS recurrence, his HIV viral load was undetectable and his CD4 count was over 900 cells/uL (CD4 percentage of 42%).

Musculoskeletal (MSK) involvement in KS is a rare manifestation of this disease. The argest series of skeletal KS in people living with HIV by Papanastasopoulos at el. showed a prevalence of only 1.1%. The radiological features of MSK-KS are generally lytic osseous lesions, but presentations may differ. Bone biopsy remains the gold standard for diagnosis, as many other infectious and neoplastic processes can mimic MSK-KS radiographically.

In the era of highly active antiretroviral therapy, people living with HIV who are diagnosed with MSK-KS appear to have a substantially improved survival rate than previously described.

Introduction

HIV-related Kaposi Sarcoma (KS) typically occurs in patients with low CD4 counts and poorly controlled HIV viral load. Musculoskeletal manifestations of KS are rare, and they are usually associated with long standing cutaneous lesions that eventually penetrate to the underlying bone [1]. Our patient represents an unusual case because his osseous lesions were multifocal and were not associated with a typical KS cutaneous lesion at the same level, but rather with a chronic plantar ulcer. The other atypical feature of this case is the fact that this patient's KS had been successfully treated four years prior. However, it relapsed despite excellent adherence to active antiretroviral therapy and very well controlled HIV.

Case report

We present here the case of a 44-year-old, African-American male with a past medical history of HIV infection on appropriate antiretroviral therapy [abacavir-dolutegravir-lamivudine], previously treated for hepatitis C, who was diagnosed with Kaposi Sarcoma (KS) four years prior and successfully treated with doxorubicin and radiation therapy. He presented to the emergency room due to worsening pain in his right foot. He reported 100% adherence to his HIV therapy. He denied any fevers or chills, but complained of foul-smelling discharge from a chronic right plantar ulcer, that has been present for 3 months. On a prior admission, patient was treated for this right foot ulcer with linezolid, ciprofloxacin and metronidazole for a total of 2 weeks, however noted no improvement, despite compliance with these broad-spectrum antibiotics.

The vital signs at the time of this presentation showed a heart rate of 87 beats/min, blood pressure 167/102 mmHg, temperature 98 °F, respiratory rate of 17 breaths/min, and oxygen saturation of 100% on room air. Physical exam revealed an erythematous, 2 x 3 cm chronic ulcer, with eschar and minimal serous drainage, on his right plant surface (Fig. 1). He also had multiple raised hyperpigmented skin lesions on bilateral inner thighs and legs.

Initial work-up showed 7550 leukocytes/uL, platelets 502,000 mm^3, erythrocyte sedimentation rate of 132 mm/hr, and a C-reactive protein of 12.4 mg/L. The white blood count differential was normal. CD4 count on presentation was 903

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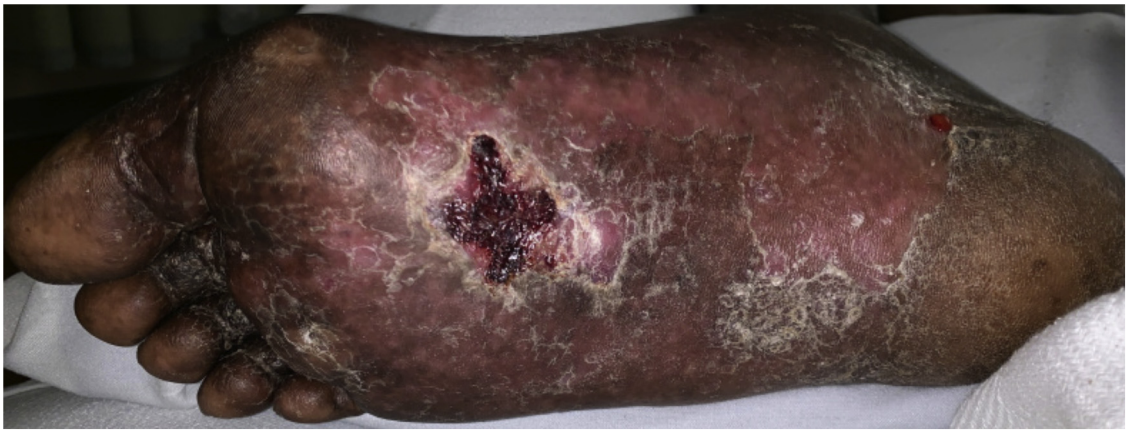

Fig. 1. Shows right plantar surface with erythematous, desquamating skin, and a 2x3 cm chronic ulcer with eschar.

cells/μL (CD4 percentage of 42%). His lowest CD4 on record was 600. Based on his records, his CD4 count had never decreased below 200 cell/μL in the past six years. His HIV viral load was undetectable on this presentation, and had remained undetectable for more than one year. Radiography of the right foot showed diffuse erosive and lytic lesions concerning for multifocal osteomyelitis (Figs. 2 and 3). Magnetic resonance imaging of the right foot done with intravenous contrast showed diffuse soft tissue swelling with mild enhancement along the plantar surface, concerning for acute cellulitis, plus multifocal intermedullary enhancing soft tissue masses with cortical erosions and soft tissue components throughout the osseous structures of the right foot (Fig. 4).

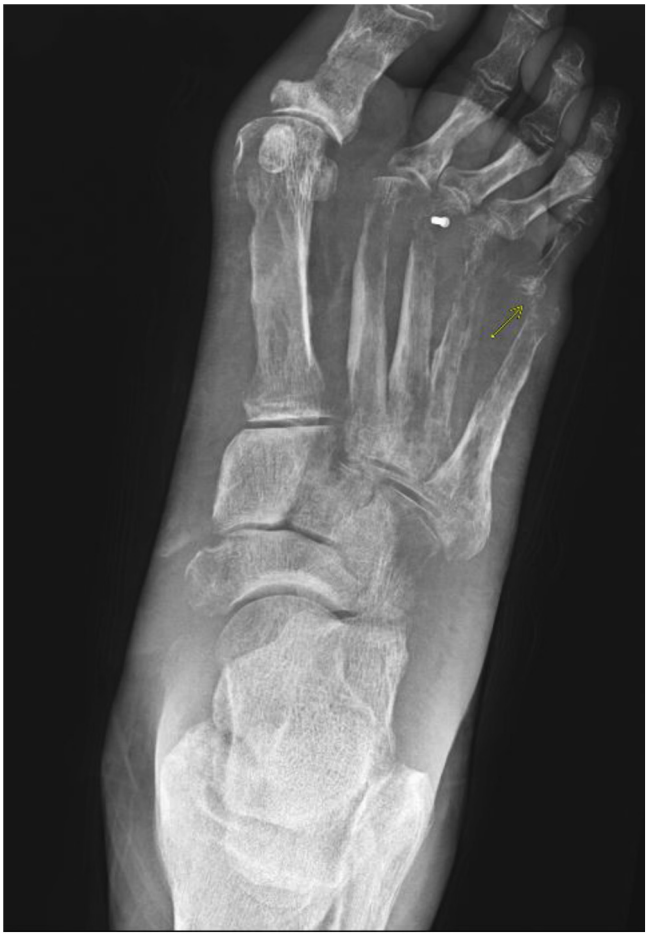

Fig. 2. X-ray of the right foot – arrow showing erosion of the fifth metatarsal head. Screw on the head of the third metatarsal from prior surgery. Osteopenia present. Soft tissue swelling is present.

Patient was initially started on broad spectrum intravenous antibiotics, which included vancomycin and piperacillin-tazobactam. Blood cultures drawn on admission remained negative. Bone biopsy was done and it revealed atypical spindle cell proliferation involving the bone, consistent with KS. Immunohistochemical tissue stain was positive for CD34 and HHV-8. Acid-fast and fungal stains on tissue were both negative. Bacterial, fungal and mycobacterial cultures of bone had no growth. Infectious disease team recommended to deescalate antibiotic regimen to oral linezolid and levofloxacin for ten days, to treat for superimposed cellulitis. A referral was made to medical oncology upon discharge, for further management of KS, along with continuation of his antiviral therapy.

Discussion

Musculoskeletal (MSK) involvement in Kaposi Sarcoma (KS) is a rare manifestation of this disease. Most cases of MSK-KS in people living with HIV (PLWH) are associated with locally aggressive, long standing cutaneous or mucosal disease that eventually penetrates underlying bone. Primary bone lesions, without concomitant skin or visceral disease are very rare [1].

The largest series of skeletal KS in PLWH by Papanastasopoulos at et. showed the prevalence of skeletal KS to be 1.1%. The same series showed that the median interval from initial KS diagnosis to MSK-KS diagnosis was 3.3 years, the median CD4+ cell count was 240/mL and fully suppressed plasma HIV viral load in the large proportion of patients at the time of MSK-KS diagnosis [2].

The radiological features of MSK-KS are lytic osseous lesions or soft-tissue masses causing osseous erosion or destruction. Sclerotic lesions have been described as well. The radiological differential diagnosis of MSK-KS is broad and includes bacillary angiomatosis, mycobacterial infections, osteomyelitis, cryptococcosis, pyogenic granuloma, hemangioendothelioma, angiosarcoma, arteriovenous malformation and lymphoma. In PLWH with known Kaposi sarcoma and new bone lesions, bone biopsy is necessary to confirm diagnosis and rule out other pathologies [3,4].

In a comprehensive literature review of MSK involvement by KS by Caponetti et al., the authors concluded that the survival following diagnosis of MSK involvement by AIDS related KS was very short, with death occurring within days or weeks. The majority of the patients in this study were not receiving antiretrovirals at the time of diagnosis and had symptomatic

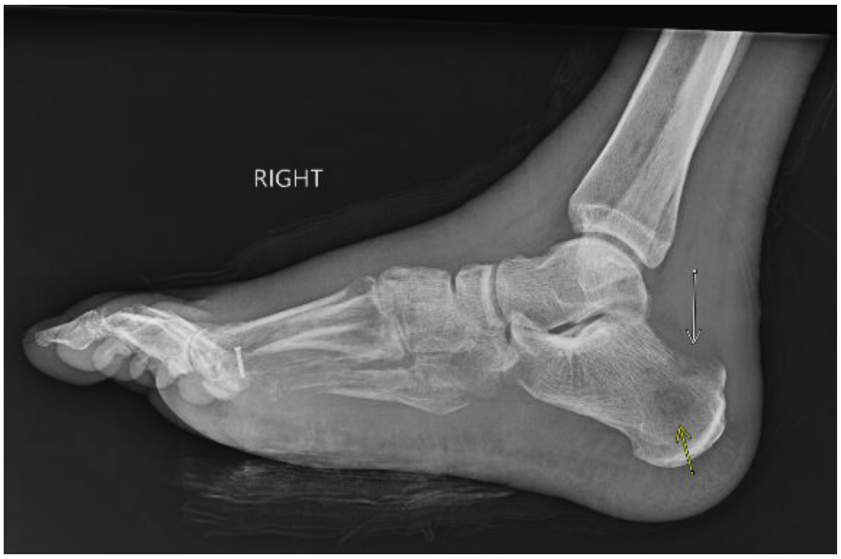

Fig. 3. X-ray of the right foot, medial aspect – arrows showing erosive lesions. Prominent tissue swelling.

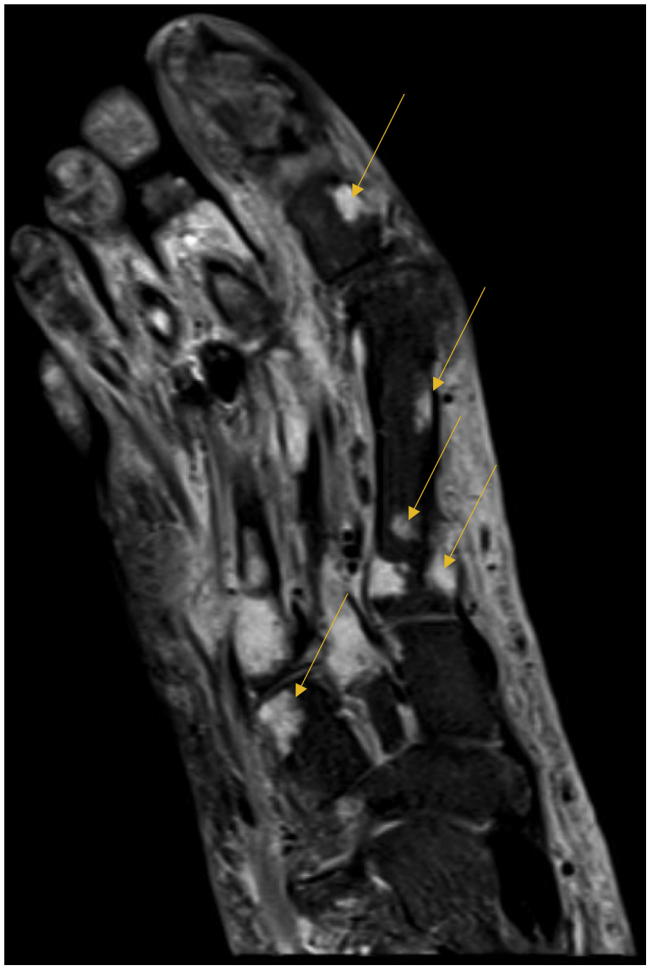

Fig. 4. MRI of the right foot – shows diffuse soft tissue swelling with mild contrast enhancement along the plantar aspect of the right foot likely representative of cellulitis. No adjacent ulceration or abscess formation. Indeterminate, multifocal intramedullary enhancing soft tissue masses with cortical erosions and soft tissue components throughout the osseous structures of the right foot.

bone disease at initial presentation [5]. Our case was different through the fact that his CD4 count was above 500 and he had an undetectable viral load, for more than one year, on his current antiretroviral regimen.

A more recent case series by Papanastasopoulos at et. showed the 5-year survival rate from diagnosis of MSK-KS to be 60%. It is not fully understood whether the improvement in survival can be attributed to a combination of antiretroviral therapy (cART) as most patients had well controlled HIV at time of the diagnosis versus due to newer chemotherapy regimens, or perhaps because of the earlier detection of MSK-KS. Most patients in this study had asymptomatic bone lesions at the time of diagnosis [2].

Conclusion

Musculoskeletal involvement represents a rare manifestation of AIDS-related KS. It can occur in patients with well-controlled HIV infection. However, this is an even rarer event. Musculoskeletal KS usually develops in the setting of longstanding, locally aggressive, cutaneous or visceral disease. Bone biopsy remains gold standard for diagnosis, as many other infectious and neoplastic processes can mimic MSK-KS radiographically. In the era of cART, PLWH diagnosed with MSK-KS appear to have a substantially improved survival rate than previously described.

Submission declaration and verification

I, Jorge Verdecia MD, corresponding author, state that submission of this article has not been published previously, that it is not under consideration for publication elsewhere, that its publication is approved by all authors and tacitly or explicitly by the responsible authorities where the work was carried out, and that, if accepted, it will not be published elsewhere in the same form, in English or in any other language, including electronically without the written consent of the copyright-holder.

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