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CASE REPORT

Kaposi Sarcoma of the Ankle Complicated by Emphysematous Osteomyelitis: A Case Report

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INTRODUCTION

Emphysematous osteomyelitis (EO) is a rare skeletal infection with a poor prognosis; mortality rate is estimated to be approximately 32%.[1] Intraosseous osteomyelitis was first described in 1981,[2] and only 34 cases have been described in the literature.[3] Recognition of this condition to facilitate prompt treatment is important for the best possible outcome.

We present a case of endemic Kaposi sarcoma in a human immunodeficiency virus–negative patient, with disease recurrence complicated by EO.

CASE PRESENTATION

A 73-year-old female with known endemic Kaposi sarcoma of the posterior left ankle diagnosed 4 years previously via skin biopsy with human herpesvirus–8 positivity presented to our institution with rapidly worsening hindfoot pain. At the time of initial diagnosis, the patient underwent 20-Gy radiotherapy and two further 8-Gy fractions over the following 2 years. She also received six cycles of 20 mg/m^2 pegylated liposomal doxorubicin. There was an excellent response and her disease remained stable until the time of her presentation 2 years later. She remained human immunodeficiency virus negative throughout her clinical encounters.

At presentation, the affected foot was extremely painful to touch. An indurated ulcer was noted at the posterior hindfoot, with the ulcer probing deep to bone; extensive associated oedema circumferentially surrounding most of the hindfoot was noted. The patient was pyrexic at 38.1°C with mild tachycardia (with a heart rate of 110 beats per minute) on admission. Blood testing returned a mild neutrophilia and a markedly elevated level of C-reactive protein. A hindfoot radiograph as part of the patient's initial workup revealed almost complete collapse of the calcaneus with extensive intraosseous gas (Figure 1).

The patient was immediately commenced on broad-spectrum antimicrobial therapy. The decision to perform an emergent amputation was made following urgent orthopaedic and oncology input on day of admission. Preoperative contrast-enhanced magnetic resonance

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Ethics Approval: The patient was treated in accordance with the Declaration of Helsinki. The patient and her son provided consent for all treatment and procedures. Separate consent was provided by the patient specifically for the publication of this case.

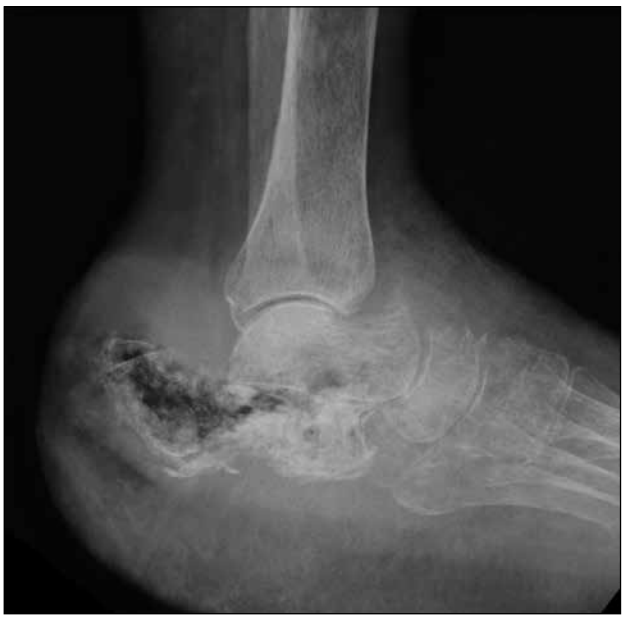

Figure 1. Radiograph showing collapse of the calcaneus with areas of intraosseous gas.

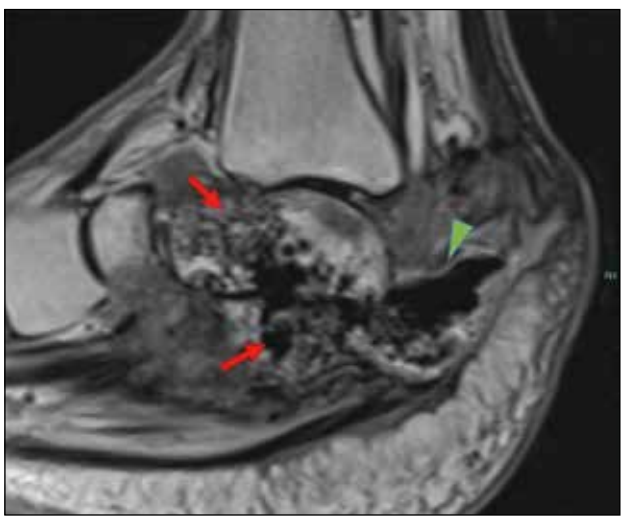

Figure 2. Sagittal T2-weighted sequence shows destruction of the calcaneus and talus, with areas of intraosseous (arrows) and extraosseous (arrowhead) signal voids, consistent with gas.

imaging was performed to aid surgical planning and assess the extent of surrounding bone and soft tissue involvement. The magnetic resonance imaging confirmed extensive destruction of the calcaneus with intraosseous gas as well as extraosseous gas surrounding the posterior aspect of the remnant calcaneus; in addition, destruction of the talus with intraosseous gas tracking into the talar head/body was noted (Figure 2). A pumice stone pattern of intraosseous gas was demonstrated

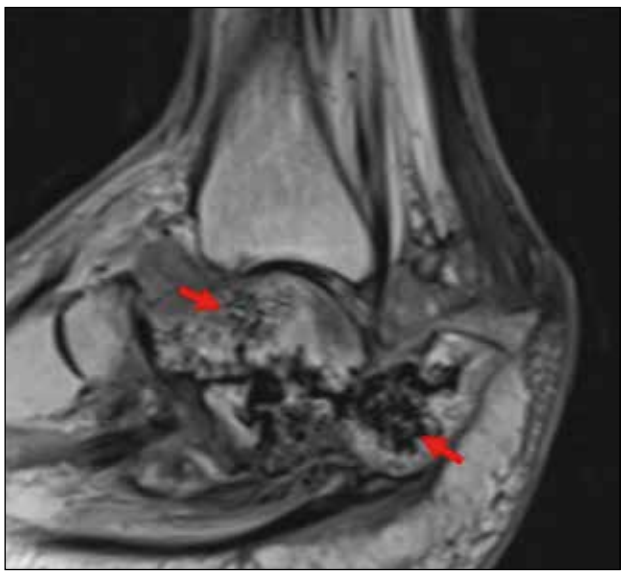

Figure 3. Sagittal T2-weighted sequence shows pumice stone pattern of intraosseous gas with clusters of sub-centimetre gas locules of irregular sizes (arrows).

(Figure 3). Following gadolinium administration, a large peripherally enhancing collection surrounding the talus and calcaneus was evident (Figure 4).

The patient subsequently underwent a below-knee amputation via a Bruckner's approach. Perioperative swabs of the ulcer and tissue sampling of the operative specimen demonstrated extensive necrosis with polymicrobial growth, including heavy growth of *Pseudomonas aeruginosa*; local recurrence of the Kaposi sarcoma was noted within the surrounding viable tissue. A subsequent postoperative fluorodeoxyglucose positron emission tomography–computed tomography showed metabolically active lung and ipsilateral inguinal nodal metastases (Figure 5).

The patient made a good postoperative recovery and was discharged from hospital on the 23rd postoperative day. A pre-discharge multidisciplinary case conference opted for a further six cycles of liposomal doxorubicin with palliative intent. The patient remains under joint outpatient care in dermatology, oncology, and palliative care.

DISCUSSION

To the best of our knowledge, there are no previously described cases of EO developing as a result of Kaposi sarcoma or its related treatment. EO is a rare and often fatal skeletal infection, with <50 reported cases in the

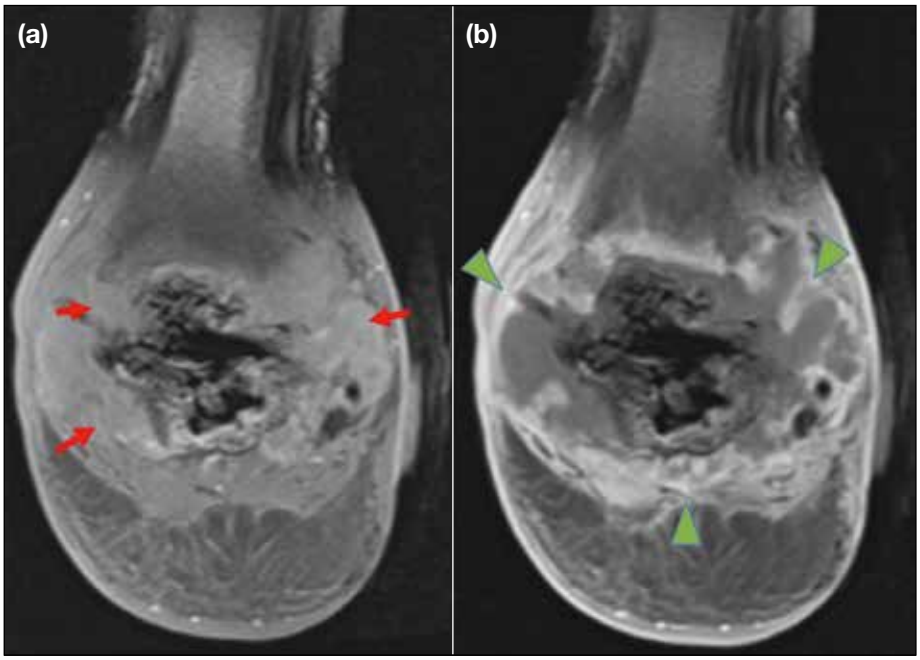

Figure 4. (a) Pre– and (b) post–gadolinium-enhanced T1-weighted fat saturated sequences show T1 signal isointensity surrounding the destructive process of the talus and calcaneus (arrows) with avid peripheral post-contrast enhancement (arrowheads), consistent with an infective collection.

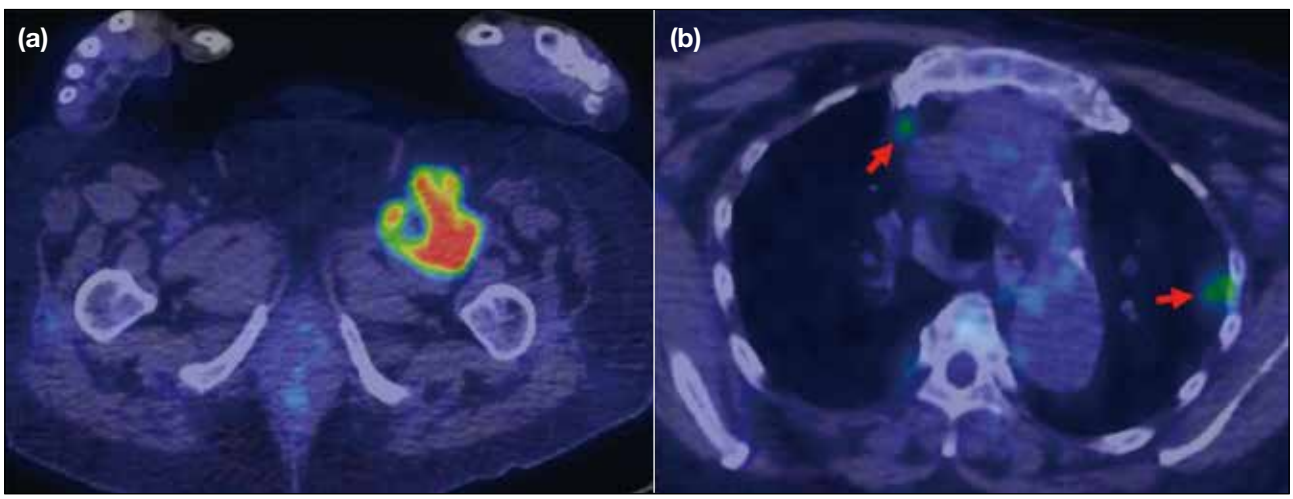

Figure 5. Fluorodeoxyglucose positron emission tomography–computed tomography fusion images show (a) large ipsilateral inguinal node demonstrating metabolic activity and (b) bilateral metabolically active lung metastasis (arrows).

literature.[1,3,4] The most common sites of involvement are the vertebrae followed by the lower extremities.[1] Haematogenous and direct contiguous spread are well-described routes of infection.[3]

The presence of intraosseous gas is a key diagnostic feature of EO.[3] A pumice stone pattern of intramedullary gas has been recently described as almost universal in EO. This imaging feature is characterised by clusters of intramedullary gas with irregular size[3]; our current case demonstrates this well. Further described secondary features include the presence of adjacent soft tissue gas and lack of cortical destruction, although these features are not universal in EO.[3] Although the presence of intraosseous gas is a prerequisite for diagnosis of EO, it should be noted that alternative causes of intraosseous gas, such as degenerative aetiologies, are much more common, although these rarely give rise to the pumice stone sign.[3] In cases of suspected EO, aggressive empirical antimicrobial treatment is required given

the prognosis; urgent surgical intervention remains the mainstay of treatment for both spinal and extremity EO.

Radiotherapy is a well-established mode of treatment for Kaposi sarcoma. As in our current case, radiotherapy may serve as the main treatment in solitary cutaneous lesions.[5] Late skin sequelae following radiotherapy are common, and the possible risk of recurrence adjacent to the radiotherapy field necessitates regular follow-up and direct inspection by an experienced dermatologist.[5] Despite regular reviews at 2 to 3 monthly intervals, our patient unfortunately presented as an emergency with an exquisitely painful foot and a rapidly progressive ulcer. We postulate that the integrity of the skin was initially compromised by rapid local disease recurrence that ultimately led to an aggressive soft tissue and bone infection via a direct route of polymicrobial seeding.

Our case highlights the importance of vigilance for rapid progression of malignancies that have been previously stable. Furthermore, we wish to highlight the utility of a cohesive multidisciplinary team in complex situations; a team of radiologist, dermatologist, clinical oncologist, orthopaedic surgeons, and infectious disease physicians were involved in the emergent management of this devastating foot infection.

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