Primary osteosarcoma of the talus

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ABSTRACT

ساركوما عظمة الكاحل ورم نادر جدا وقد نُشرت حالات قليلة جدا في الدوريات الطبية. ونسجل حالة مريض يبلغ من العمر ٣٣ عاماً كان يشكو من ألم وتورم في كاحله الأيسر، وقد أجريت له العديد من الفحوصات الاشعاعيه والتي بينت وجود انحلال عظمي في الجزء الخلفي من عظم الكاحل متصاحبا مع تكون عظمي جديد يبرز من الجزء الخلفي الداخلي لعظم الكاحل. و أوضحت أشعة الصدر وجود العديد من الكتل الكروية منتشرة في كلا الرئتين، بعض هذه الكتل بينت تكلسات مما يدل على أنها انتشار ثانوي من ورم عظمي. أخذت عينه من عظم الكاحل، والتي بينت بالفحص المخبري النسيجي أنها ساركوما عظمية أولية.

Osteosarcoma (OS) of the talus is extremely rare, and few cases have been reported in the literature. We present a case of a 33-year-old male with painful swelling of his left ankle joint. He underwent several radiological diagnostic modalities that showed osteolytic lesion in the posterior aspect of the left talus associated with new bone formation projecting from the posterior-medial aspect of that bone. His chest x-ray showed multiple rounded lung metastases, some of them showed calcifications. Open biopsy was performed. The histopathology confirmed the diagnosis of osteoblastic OS of the talus.

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Osteosarcoma (OS) is the most common primary bone malignancy. In most cases, this tumor occurs in the metaphysis of long bones such as femur, tibia, or humerus.[1] The calcaneus and metatarsals are the favorite sites of this tumor in the foot.[2] The OS of the talus is extremely rare, and few cases have been reported in the literature.[1-3] Its clinical findings are usually not typical, and can be easily misdiagnosed, resulting in a delay in proper treatment.[3] We aim to raise awareness on another rare case of primary OS of the talus, with multiple lung metastases and describe the imaging features, and confirmed the diagnosis by histopathological examination.

Case Report. A 33-year-old male presented to the orthopedic clinic due to increasing pain and swelling in his left ankle joint over a period of 8 months. His medical history was not remarkable. There was no apparent tumor mass. The x-ray of the right ankle joint was normal, and he was treated conservatively. Four months later, his physical examination showed tenderness and a hard swelling in the posterior aspect of the left ankle. Plain x-ray of the left ankle joint (**Figure 1a**) showed an ill-defined osteolytic lesion in the posterior aspect of the talus associated with new bone formation, and bony outgrowth projecting from the posterior-medial aspect of that bone. His chest x-ray showed bilateral multiple lung metastases (**Figure 1b**). A computed tomography (CT) scan of the ankle joint confirmed the presence of an osteolytic bony lesion (**Figure 2**) involving the talus with new bone formation, and bony outgrowth projecting from the posterio-medial aspect of the talus with no periosteal reaction and no invasion of the adjacent joints. Chest CT scan showed bilateral multiple rounded lung metastases, and some of them showed calcifications (**Figure 3**). His routine laboratory investigations were within normal limits, except for an increased leukocytic count and an elevated alkaline phosphatase. Open biopsy was performed. Histopathology (**Figure 4**) showed a tumor composed of spindle cells arranged in interlacing fascicles with nuclear pleomorphism. There were areas of mitosis and necrosis, and extensive amounts of osteoid and calcified bone matrix. The immunostaining showed immuno activity for Vimentin. There was no reactivity for endomysial antibodies, creatinine kinase, or carcinoembryonic antigen. The final diagnosis was consistent with osteoblastic OS, moderately differentiated (grade II) involving the talus with multiple lung metastases. The patient was referred to a specialized oncology center for chemotherapy.

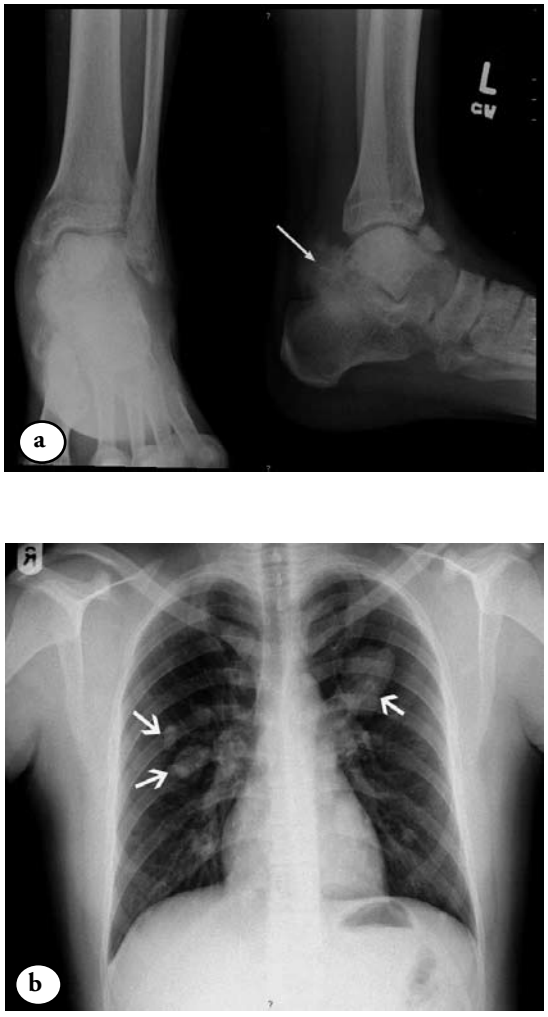

Figure 1 - Radiological results of **a**) antero posterior and lateral views of the left ankle joint showing an ill-defined osteolytic lesion at the posterio-medial aspect of the talus with bony outgrowth (arrow), and **b**) chest x-ray showing bilateral multiple lung metastases (arrows).

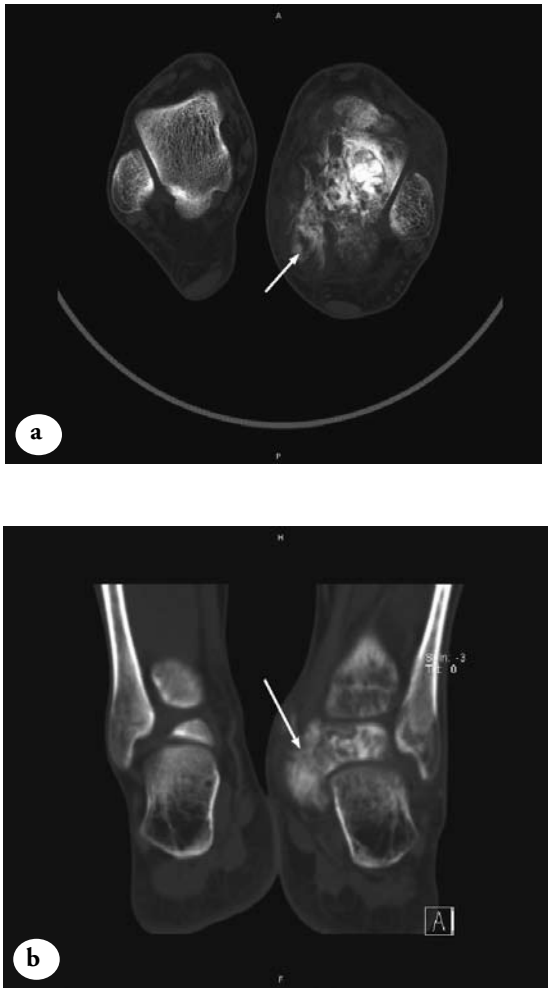

Figure 2 - Computed tomography scan of both ankle joints bone window of **a**) axial section, and **b**) coronal reformatted showing a destructive bony lesion involving the left talus with new bone formation and bony outgrowth projecting from the posterio-medial aspect of the left talus (arrow) and preserved adjacent joints.

Discussion. The reported incidence of OS of the foot is very low (1%).[2] The calcaneus and metatarsals are favorite sites of this tumor in the foot. However, OS of the talus is extremely rare and few cases have been reported in the literature.[1,4] The talus is essentially a rare site of primary bone tumor. According to literature review, bone tumors originating in the talus were osteochondroma, chondroblastoma, giant cell tumor, osteoblastoma, aneurysmal bone cyst, Ewing's sarcoma, angiosarcoma, solitary myeloma, and metastatic tumor. Amnii and Colacecchi[5] reported the first case of OS involving the talus in 1980. In our patient, and the case reported by Amnii and Colacecchi,[5] the radiographic findings of ill-defined bone destruction with new bone formation suggested the osteogenic malignant nature of the lesion. Based on these findings, the radiological diagnosis at first presentation for the current patient was OS instead of the other bone tumors more commonly affecting the talus. The rarity of OS affecting the tarsal bones may lead physicians to misdiagnose or delay proper treatment. In addition, OS of the talus shows no typical periosteal reactions such as Codman's triangle and spicules that are present in OS of long bones. The radiological investigations including plain x-ray and CT scan can be useful tools regarding site, size, extent, degree of bone destruction, and degree of invasion by the bony tumor. In OS, plain films were the primary

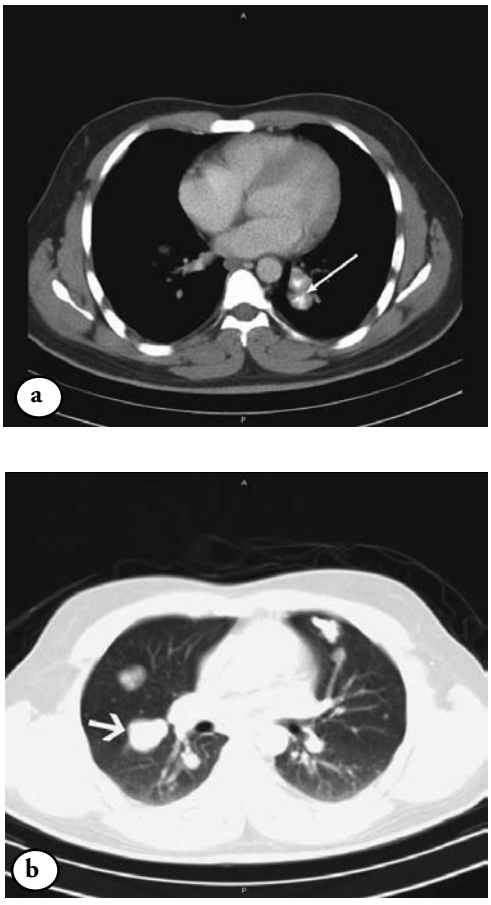

Figure 3 - Computed tomography scan of **a**) chest soft tissue, and **b**) lung windows showing bilateral multiple lung metastatic nodules with calcifications within the nodule (arrow).

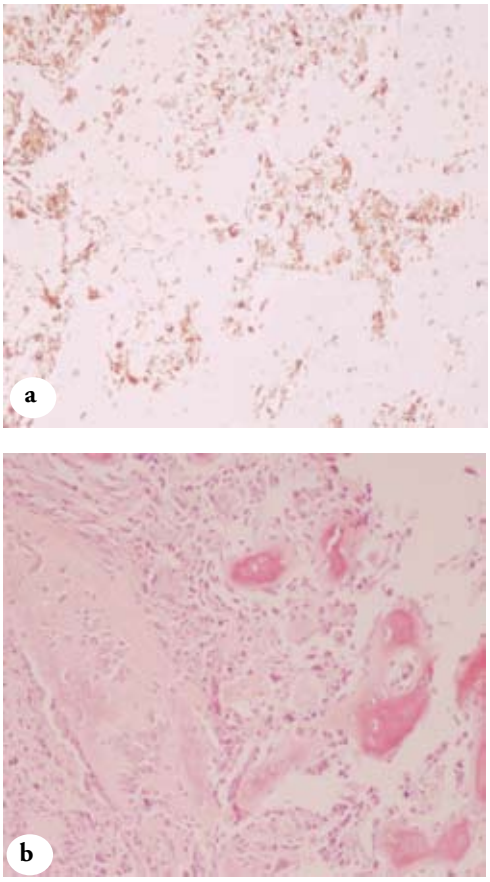

Figure 4 - Histopathology with hematoxylin and eosin staining showing **a**) tumor composed of spindle cells with pleomorphism, mitosis, extensive amounts of osteoid and calcified bone matrix, and **b**) immunostaining showing immunoreactivity to Vimentin.

radiologic tools for the investigation. Bony changes may show lytic or sclerotic osteoid bone matrix in the lesion.[6] A CT scan was able to show new bone formation in the soft-tissue mass that was not seen on plain films.[6] An important feature on CT scans is the spatial distribution of areas of mineralization, which is greatest at the center of the lesion and least at the periphery. The main value of the radionuclide bone scans is to detect metastatic or multifocal bone disease.[6] Approximately 34% of patients with OS have metastasis at time of presentation.[7] The lungs should be followed at regular intervals with plain chest radiographs and CT scans for early detection of lung metastasis. The prognosis remains poor, and unfortunately the 5 year survival is around 15% with a median of 12 months.[7]

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