Imaging of a Marjolin's Ulcer: A Case Report[1]

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A Marjolin's ulcer refers to malignancies that developed in chronic venous ulcers, scars, or sinuses. We report three-dimensional computed tomography (CT), magnetic resonance imaging (MRI), and positron emission tomography (PET)-CT findings in a patient who developed skin cancer from a chronic leg ulcer. Although rare, on MR, a Marjolin's ulcer should be considered when a well-enhanced soft-tissue mass with a broad based skin ulcer shows a mass effect and invasion of the adjacent bone. CT angiography and PET-CT complement MRI for evaluating the nature of Marjolin's ulcers and may provide essential anatomical information, enabling the physician to design the optimal surgical approach or determining cancer staging.

Index words : Magnetic Resonance Imaging
Tomography, X-Ray Computed
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The term "Marjolin's ulcer" refers to malignancies that developed in chronic venous ulcers, scars, or sinuses (1, 2). These malignancies also arise from various scars, including chronic ulcerations, inflammation, and fistulas after a long period of latency. Malignant transformation takes approximately 35 years. The incidence of malignant skin tumors that developed from scar tissue is 0.1–2.5%. Burn scars are the most common lesion causing this malignancy (3). The most common malignancy that arises from a Marjolin's ulcer is squamous cell carcinoma, whereas basal cell carcinomas are rare. The malignancy is frequently multiple in the floor of an ulcer (3).

The pathogenesis of a Marjolin's ulcer is controversial (1, 3). Ulcer osteomas develop as a result of the heaping up of periosteum and associated subperiosteal sclerosis, and were seen as a knob-like mass protruding from the bone surface (1). These masses are rare in patients with chronic ulcers (1); however, they are frequently seen in patients with tropical ulcers, occurring in form of cutaneous leishmaniasis, which is a skin infection caused by a single-celled parasite that is transmitted by sandfly bites (4).

A previous report described gadopentetate dimeglumine-enhanced magnetic resonance imaging (MRI) as a useful tool for evaluating a Marjolin's ulcer (2). A multidisciplinary approach using three-dimensional computed tomography (CT) and positron emission tomography

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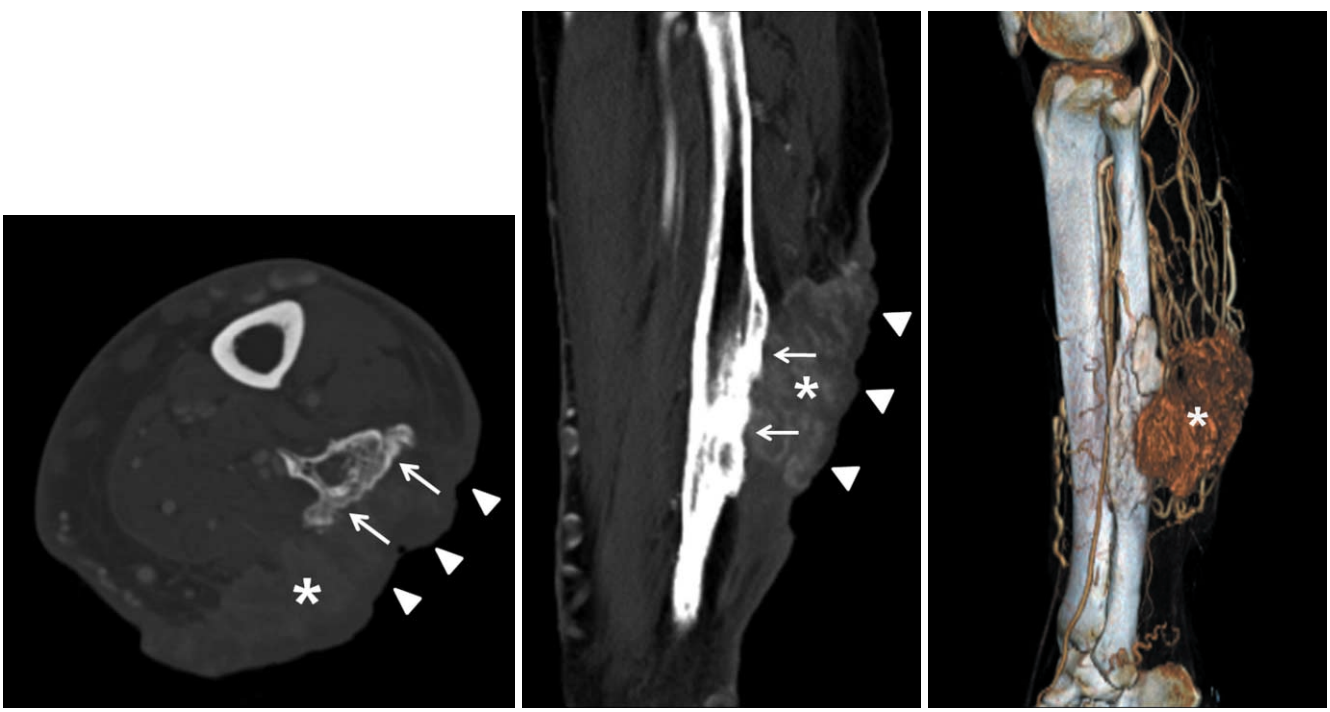
(PET)-CT is essential for the imaging workup of malignant cancers for metastatic disease. To our knowledge, the presentation of imaging features of a Marjolin's ulcer as a soft-tissue mass with an ulcer osteoma have seldom been reported in the radiological literature. Here, we report on a case of a Marjolin's ulcer, with an emphasis on a multimodality approach including 3D CT, MRI, and PET-CT.

Case Report

A 57-year-old woman presented with a 2-month history of a progressively worsening swelling on her left leg and discharge from a scar. She had suffered a flame burn injury 10 years earlier. Subsequently, there had been no marked change in the burn scar on her left leg. However, over the 6 months before her presentation, a mass in the scar had progressively increased in size, and was associated with bloody discharge. Laboratory results were within normal limits.

Radiography of the left lower leg showed an ulcer osteoma cortical thickening on the lateral aspect of left

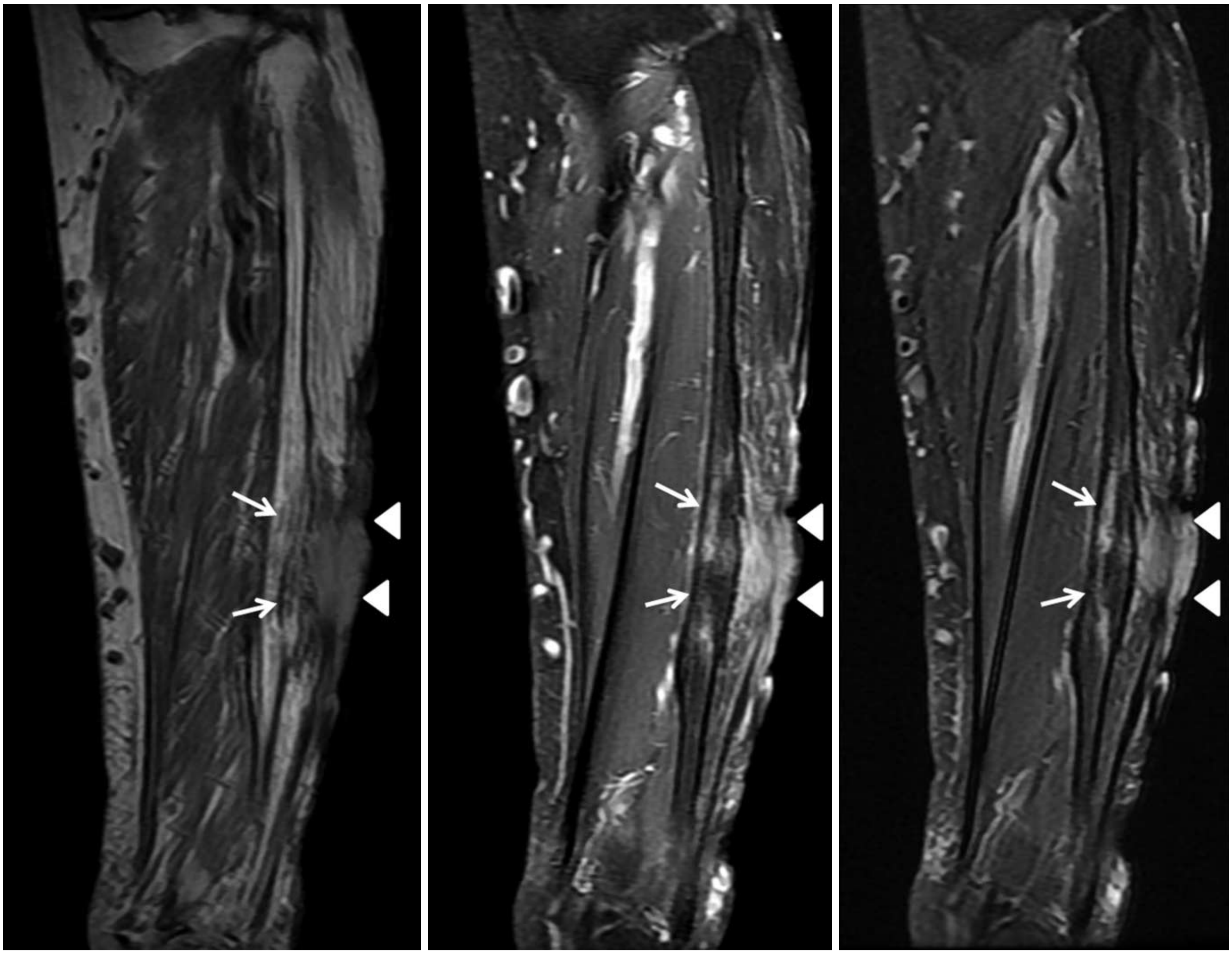
Fig. 1. Anteroposterior radiograph of the left lower leg shows cortical thickening of the fibular diaphysis (arrowheads) beneath the skin ulcer (arrow). Note the cortical hypertrophy on the opposite side of the skin ulcer.



A B C

Fig. 2. Computed tomography.
A. Axial CT with bone setting reveals cortical hypertrophy of the left lateral fibula (arrows) extending anteroposteriorly parallel to the skin ulcer (arrowheads). A soft tissue mass (asterisk) is also seen between the skin ulcer and fibula.
B. Sagittal reformatted contrast-enhanced CT shows a thickened cortex (arrows), which is extrinsically eroded by a soft-tissue mass (asterisk) with a broad base on the skin ulcer (arrowheads).
C. CT angiography with color-coded volume-rendering demonstrates a well-enhanced soft-tissue mass (asterisk) surrounded by numerous supplying arteries.

fibula, adjacent to the skin ulcer (Fig. 1). Contrast-enhanced CT demonstrated cortical erosion on the ulcer osteoma and a well-enhanced soft-tissue mass with a broad base in the skin ulcer (Figs. 2A, B). CT angiography showed a highly vascular mass supplied by multiple arterial feeders (Fig. 2C). MRI was performed with a 3.0 T MR scanner (Signa HDxt; GE healthcare, Milwaukee, Wis, USA). The soft-tissue mass abutting



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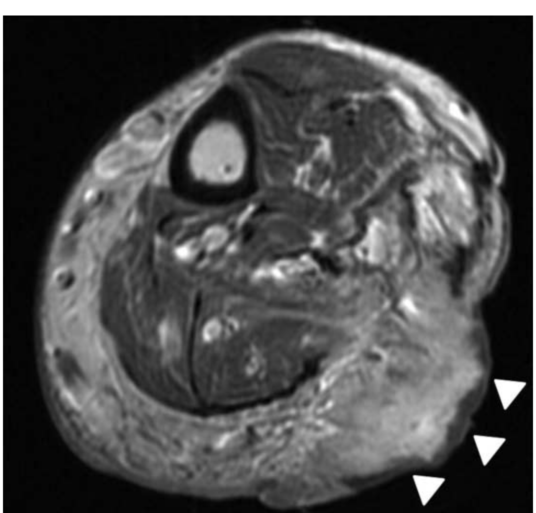

D

Fig. 3. Magnetic resonance imaging.
A-C. The soft tissue mass (arrowheads) shows slightly greater signal intensity than muscle on (A) coronal T1weighted images [repetition time (TR) 1466, echo time (TE) 15], (B) high signal intensity on fat-saturated T2-weighted images (TR 3550, TE 65), and (C) strong enhancement on contrast enhanced fat saturated T1-weighted images (TR 833,TE 13). Note the cortical involvement of the soft tissue mass and abnormal signal intensity lesions in the bone marrow (arrows).
D. The axial contrast-enhanced T1-weighted image (TR 900, TE 11) shows a highly homogenous enhanced mass bulging out from the skin scar.

the skin ulcer showed low signal intensity on T1-weighted images, high signal intensity on fat-saturated T2-weighted images, and strong enhancement on contrast-enhanced T1-weighted images (Figs. 3A–D). In addition, fat saturated T2 weighted images showed high signal intensity lesions in bone marrow surrounded by the cortical hypertrophy which showed a patchy enhancement on contrast enhanced fat saturated T1 weighted images. PET-CT showed intense fluorodeoxyglucose (FDG) uptake (standardized uptake value, SUV = 11.2) in the mass, but with no abnormal uptake in the bone marrow adjacent to the skin ulcer (Fig. 4). An excisional biopsy of the soft-tissue mass was performed and the microscopic analysis revealed squamous cell cancer in the soft tissue mass and lymph nodes (Fig. 5). After neoadjuvant chemotherapy, the patient underwent an above-knee amputation based on the CT and MRI findings. The pathological specimen obtained at amputation confirmed tumor involvement in the bone marrow which corresponded to the area with high signal intensity on a T2 weighted image and strong enhancement on a contrast enhanced T1 weighted image (Fig. 3).

Discussion

An ulcer-osteoma was first described in patients with tropical ulcers from West Africa (1). Periosteal reaction is the earliest radiological finding and is initially present in the bone beneath the ulcer only. Further, the periosteal reaction has a fusiform shape, but occasionally looks like a sunburst. This periosteal new bone eventually fuses with the original cortex, which results in the thickening of the sclerotic cortex by more than 2.5 cm, giving rise to the classical 'ivory ulcer osteoma.' The periosteal reaction may eventually involve the entire circumference of the long bone shaft (4).

In our case, the fusiform cortical hypertrophy beneath the skin ulcer was clearly demonstrated on CT. Karasick et al. (5), whom described two types of periosteal responses to chronic leg ulcers: a solid organized type that forms an ulcer osteoma over time and a lamellar nodular type that is often associated with osteomyelitis. Karasick et al. further indicated that patients with both types of periosteal reaction usually had associated peripheral vascular disease, intravenous drug abuse, sickle cell disease, or neurologic impairment (5). Unlike these previous reports, an ulcer-osteoma adjacent to a skin ulcer occurred in our patient, who had no clinical or laboratory finding of the underlying diseases previously associated with ulcer osteomas (5).

Chiang et al. (2) reported that contrast-enhanced MRI is the imaging technique of choice for assessing a Marjolin's ulcer, because MRI is superior at characterizing the nature and extent of the ulcer (1). In our study, a highly enhanced mass in the skin ulcer site was clearly seen on contrast-enhanced MRI, allowing for the assessment of the margin and depth of the tumor.

Computed tomography has an advantage over MRI for detecting cortical destruction, which helps to determine the relationship between the soft-tissue tumor and adjacent bone (6). With the advent of multi-detector

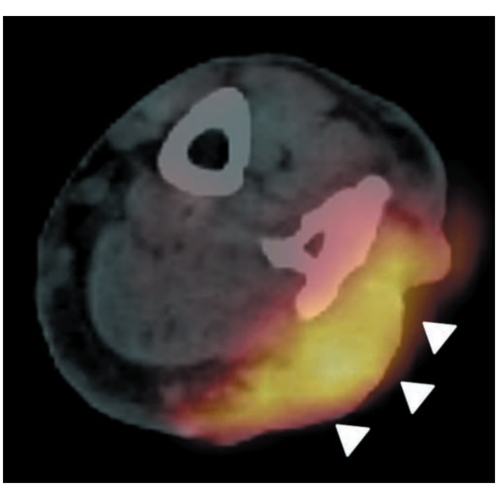

Fig. 4. PET/CT.
Transverse PET/CT shows intense FDG uptake by the soft tissue mass (arrowheads), which is seen as a highly enhanced mass on CT and MR images. There was no abnormal FDG uptake in the bone marrow where abnormal signal intensity was seen on MRI.

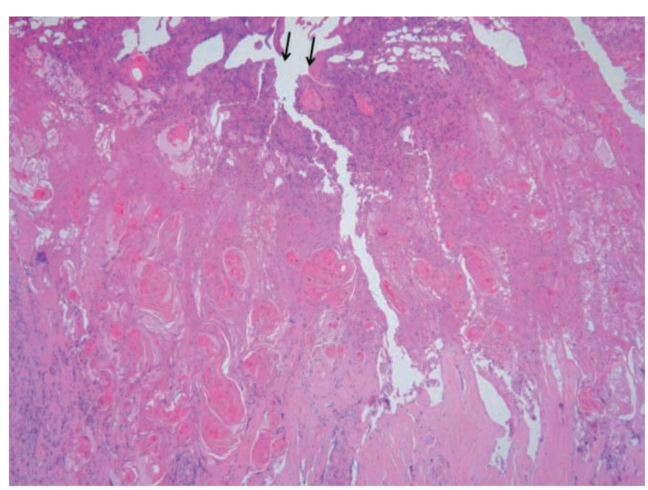

Fig. 5. Microscopically, nests of atypical squamous cells with keratinization spread from the ulcerated epidermis (arrows) into the deep dermis (hematoxylin and eosin staining; original magnification, × 40).

(MD) CT, the use of three-dimensional (3D) CT provides better anatomical resolution for delineating the nature and extent of the bony involvement for preoperative planning (6). As seen in our case, multiplanar CT images with 3D reconstruction demonstrated the extent of the cortical erosion caused by direct extension of the tumor, and the details of the ulcer-osteoma. Furthermore, on CT angiography with volume rendering, multiple arterial feeders were seen supplying the tumor mass; this may help in identifying malignant tumors.

Recently, PET-CT imaging has increasingly been used for the diagnosis and staging of tumors. Studies indicate that 18F-FDG-PET-CT reliably differentiated malignant soft-tissue and bone tumors from benign ones (7, 8), although there was considerable overlap in the maximum SUV value between benign and malignant tumors (9). Shin et al. (7) reported that the sensitivity, specificity, and diagnostic accuracy of 18FFDG-PET-CT were 80%, 68.4%, and 75%, respectively, for soft-tissue tumors with a maximum SUV cutoff of 3.8. In our case, the soft tissue mass had a SUV approximately three times greater than 3.8, strongly suggesting that the tumor was malignant. However, PET-CT did not demonstrate tumor involvement of the bone marrow just below the area of cortical destruction, which was seen in MRI images.

Wide local excision (surgical margin of at least 2 cm) combined with skin grafting is the treatment of choice for a Marjolin's ulcer (10). Amputation is considered only when the lesions involve a joint, invade the limb bones, or there is deep extensive local invasion (2). The roles of routine regional lymph node dissection and irradiation remain controversial (2).

In conclusion, although rare, a Marjolin's ulcer should be considered when a well-enhanced and broad based soft-tissue mass on a skin defect shows mass effects and invasion to an adjacent ulcer osteoma. CT angiography with volume rendering and PET-CT complement MRI when evaluating the nature of Marjolin's ulcer, and may provide essential anatomical information for the surgeon when designing the optimal surgical approach or determination of cancer staging.

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Marjolin's Ulcer의 영상 소견: 증례 보고[1]

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A Marjolin's ulcer는 만성 정맥성 궤양, 흉터, 동(sinus)에서 발생한 악성 종양을 말한다. 저자들은 만성 하지 궤양에 발생한 피부암 환자의 3차원 CT, MR, PET-CT 소견을 보고한다. A Marjolin's ulcer는 드물지만, 조영 증강을 보이는 연부조직 종괴가 피부 궤양에 넓은 기저부를 형성하고 종괴 효과를 보이면서 궤양성 골종을 침범했을 경우 A Marjolin's ulcer를 의심해야 한다. CT angiography와 PET-CT 영상을 통해서 A Marjolin's ulcer의 특성을 평가하는데 MR영상을 보완할 수 있었고 필수적인 해부학적 정보를 제공 함으로써 효과적인 외과적 계획 수립과 종양의 병기 결정을 도움을 줄 수 있었다.