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CASE REPORT

Marjolin's Ulcer

Radiographic and magnetic resonance appearances in two cases

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قرحة مارجولين

المظاهر الشعاعية وصفات الرنين المغناطيسي في حالتين

سخبال سوهني ، راجيف جين، أنوبام كاكاريا براديب جوبرا

الملخص: قرحة مارجولين قرحة خبيثة تحصل في تقرحات الوريد المزمنة والندب والحروق والجروح المزمنة . يعطي التصوير الشعاعي معلومات مهمة حول تآكل العظم والتفاعل السمحاقي . بينما يبين التصوير بالرنين المغناطيسي تفاصيل الأنسجة الرخوة بشكل دقيق . مثل انتشار الورم وعمقه وحافاته وأي اصابة في قشرة العظم أو نقيه. أو اصابة الأعصاب والأوعية الدموية المجاورة. ندرج هنا حالتين لقرحة مارجولين مع وصف للتصوير بالأشعة والرنين المغناطيسي.

مفتاح الكلمات: قرحة مارجولين، قرحة جروح الساق المزمنة، تصوير الرنين المغناطيسي، تقرير حالة ،عمان.

ABSTRACT: Marjolin's ulcers are malignancies that arise in chronic venous ulcers, scars, burns, long standing wounds or sinuses. Radiography provides important information regarding bone destruction and periosteal reaction, and magnetic resonance (MR) imaging provides excellent soft tissue detail, like tumour extent, depth, margins, any underlying bone cortical or marrow involvement, or involvement of adjacent neuro-vascular structures. We report two cases of Marjolin's ulcer and describe their radiographic and MR appearances.

Keywords: Marjolin's Ulcer; Chronic wound; Leg ulcers; Magnetic resonance imaging; Case report; Oman

Marjolin's ulcers are rare malignancies that arise at sites of chronic non-healing wounds and scar wounds. The precise mechanism by which chronic ulcers develop malignancy is unknown. The commonest type of carcinoma arising in the Marjolin's ulcer is a squamous cell carcinoma, followed by basal cell carcinoma. We report two cases of Marjolin's ulcer along with radiographic and MRI findings

Case One

A 70 year old man developed a chronic non-healing ulcer on his right leg following a childhood trauma and presented at Sultan Qaboos University Hospital (SQUH), Oman. The ulcer had become worse during the previous two years. It had started increasing in size, was oozing blood and had become painful. On examination, he had a large fungating ulcerative mass over the antero-lateral aspect of his right lower leg and right inguinal lymph nodes were palpable. The remainder of his physical examination was unremarkable. A punch biopsy of the ulcer revealed well differentiated squamous cell carcinoma. Radiographs [Figure 1] and a magnetic resonance imaging scan (MRI) [Figure 2a, 2b & 2c] confirmed the findings of a large polypoidal soft-tissue mass invading the underlying tibial cortex and marrow. The patient was advised amputation and block dissection which he refused and left the hospital against medical advice.

Case Two

An 85 year old man developed an infective ulcer over a chronic wound on his left lower leg which he had sustained 10 years previously. Despite local and systemic treatment the ulcer was becoming larger and painful. On presentation at SQUH, the examination showed a large, polypoidal, fungating ulcerative mass, measuring 7cm x 7cm located on the anterior surface of the tibia. A biopsy of the ulcer was positive for squamous cell carcinoma. The

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Figure 1: (Patient 1) A radiograph of right lower leg reveals a lobulated soft tissue mass over the anterolateral surface of the leg. The underlying tibia shows lamellated, thick periosteal reaction and an underlying ill defined lucent lesion in the medullary shaft.

rest of his physical examination was unremarkable. No metastatic lesions were found on the computed tomography (CT) scan. A Tc 99m-MDP bone scan revealed abnormal increased tracer uptake at the local site in the tibia with no other abnormal uptake in the rest of the skeleton.

The radiograph [Figure 3], a bone scan [Figure 4] and MRI examinations [Figure 5a, 5b & 5c] were performed which revealed a large infiltrative

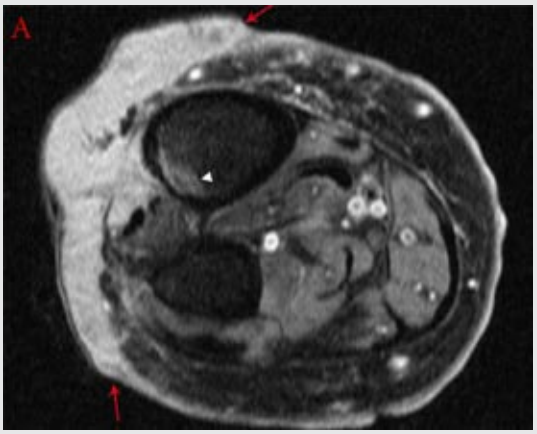

Figure 2a: (Patient 1) Transverse T2-weighted magnetic resonance imaging (MRI) scan shows a lobulated, pretibial skin and subcutaneous infiltrative soft tissue mass (arrows) extending posteriorly, invading the muscles of the lateral compartment and involving the underlying tibial cortex and medulla (arrowhead).

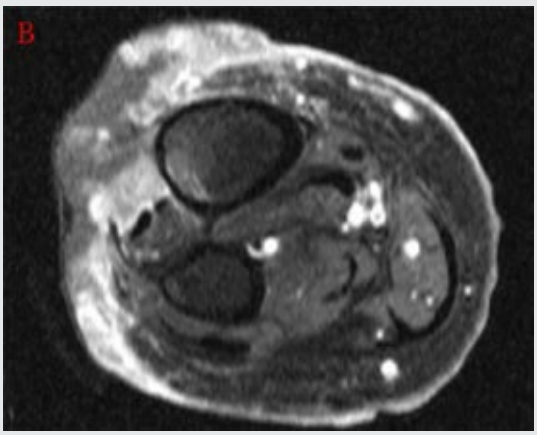

Figure 2b: Contrast enhanced transverse TI-fat saturated image shows a circumferential mass with patchy intense enhancement and necrotic areas. The bone marrow of the tibia shows a focal moderately enhancing area beneath the mass.

Figure 2c: Contrast enhanced transverse TI-fat saturated image demonstrates the longitudinal extent of the mass and infiltration into the bone.

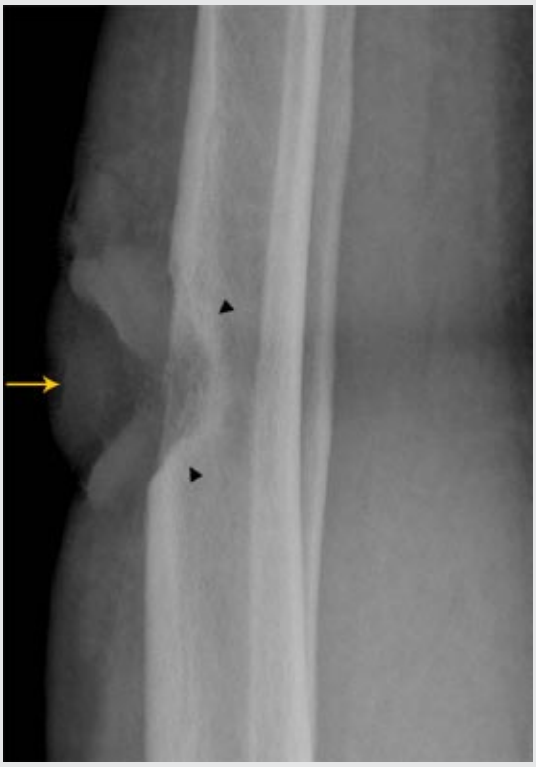

Figure 3: (Patient 2) A radiograph of the left lower leg revealed a large lobulated soft tissue mass with a central ulcerated area (arrow) encroaching on the on the anterior tibial shaft and causing a punched out osteolytic defect with sclerotic margins (arrowheads).

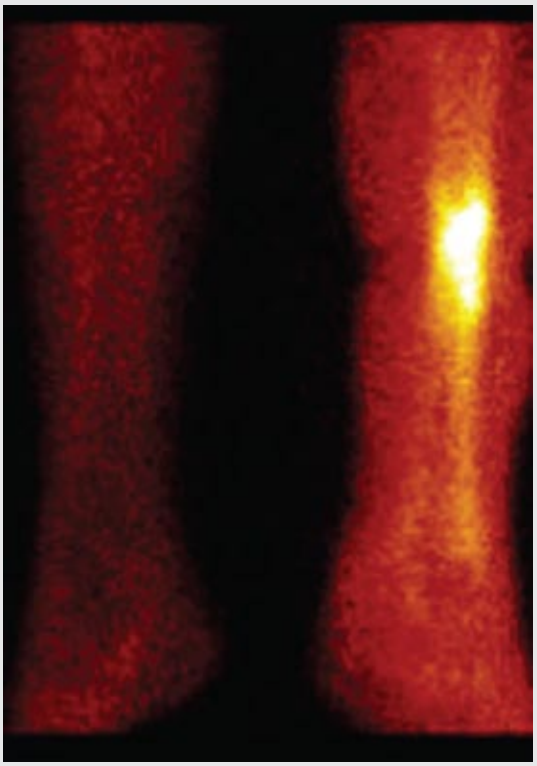

Figure 4: (Patient 2) Bone scan showed a region of intense uptake in the mid-leg

superficial mass extending through the underlying tibial cortex into the medullary cavity. The patient was advised amputation which he subsequently refused.

Discussion

Malignancies that arise in chronic venous ulcers, chronic injuries, scars, burns, chronic osteomyelitis or sinuses are referred to as Marjolin's ulcers. Marjolin's ulcer is an extremely rare condition which is reported to develop in approximately 0.05%[1] to 0.05%,[2] of long-standing pressure ulcers in patients with spinal cord injuries. A French surgeon, Jean-Nicolas Marjolin, at the University of Paris described the occurrence of ulcerating lesions within scar tissue in 1828;[3] however, he did not identify the warty ulcers he described as malignant. Two years later it was Dupuytren[4] who noted that these lesions were malignant. The commonest type of carcinoma arising from Marjolin's ulcer is squamous cell carcinoma, followed by basal cell carcinoma. The exact mechanism by which chronic ulcers (wounds) develop malignancy is not known. A variety of causes including chronic irritation and infection (with resulting degeneration and regeneration); decreased vascularity and a weakened epithelium, and elevated expression of proto-oncogenes,[5] have been suggested for the susceptibility of chronic wounds to malignant transformation. Inflammation, ulceration, and repeated trauma, especially in flexion creases, over many years may provide enough chronic irritation to promote malignant change.[6]

The pathogenesis of Marjolin's ulcer remains unclear and controversial. Early theories suggested that cellular mutations as a result of toxins release by damaged, ischaemic and nutritionally deficient tissues are responsible for neoplastic change. [7] Although the latent period between the original injury and the development of Marjolin's ulcer has been documented as being as being as long as 30 years,[8] Thio *et al*.[9] have reported a case of Marjolin's degeneration that developed in an ulcer only 18 months after the initial injury.

Bone destruction is the most important radiological finding for the surgeon. It was found in 20 out of 21 patients described by Smith *et al*.[5]

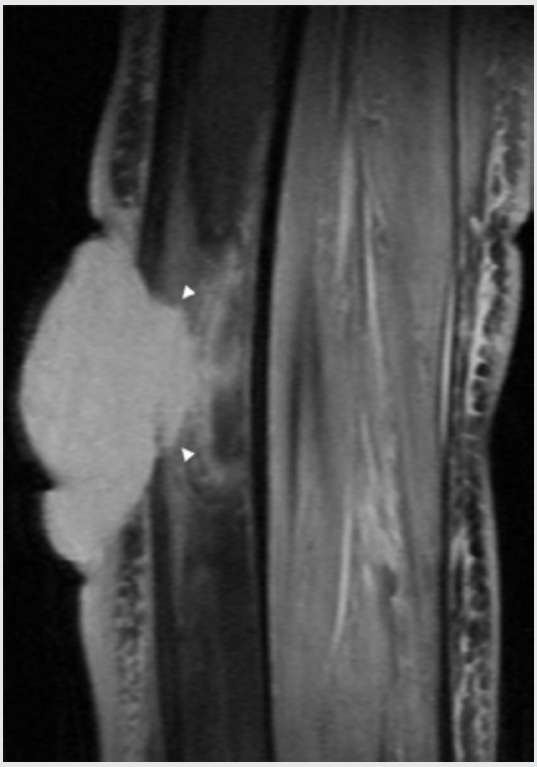

Fig5a: (Patient 2) Sagittal PD-weighted image shows a large lobulated mass with infiltration into and destruction of the cortex with extension into the medullary cavity (arrowheads)

In only one patient in their study, bone destruction could not be seen due to sub-optimal quality of the radiograph. Both our patients demonstrated bone destruction on radiographs along with periosteal reaction.

The MRI scan had the advantage of demonstrating excellent soft tissue detail, the extent of the ulcer, its margins, the extension of tumour into cortex and bone marrow, the periosteal reaction and the involvement of surrounding structures.

MRI scanning is superior to CT as it demonstrates the extent of medullary bone, soft tissue and neuro-vascular involvement. Most frequently, both T1 and T2 weighted images are needed to characterise the lesions. T2 weighted images are best suited for soft tissue characterisation and T1 weighted images for distinction between marrow and tumour. Short-tau inversion-recovery (STIR) sequences are useful for detection of subtle marrow or soft tissue lesions. To date, gadolinium has not significantly improved the histological accuracy of MRI except for differentiating solid from cystic lesions and identifying areas of necrosis.[10,11] Bone scans help to identify areas of increased radioisotope uptake.

Conclusion

To conclude, Marjolin's ulcer is a rare, but important entity, which may be preventable by the early treatment of non-healing ulcers. These ulcers should be followed up with frequent biopsies and imaging evaluation to detect or exclude infiltration of adjacent tissues by an undetected deeper focus of malignancy. The ideal imaging techniques for evaluation of soft-tissues and infiltration of underlying bone and critical neurovascular structures is MRI.

Contrast enhanced transverse T1-fat saturated

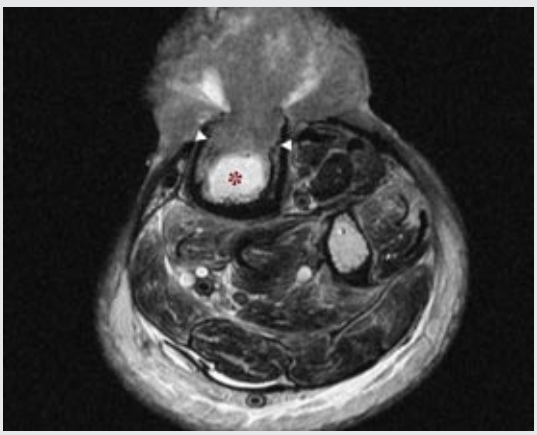

Figure 5b: (Patient 2) Transverse T2-weighted image demonstrates the infiltration the cortex (arrowheads) and the medullary cavity (*) by the mass. Predominantly low signal intensity of the mass reflects the fibrotic nature of squamous cell carcinoma

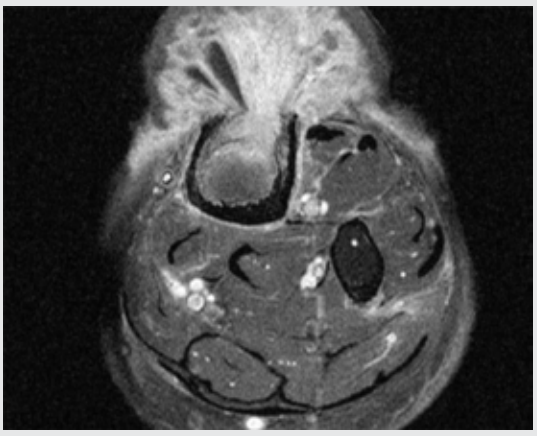

Figure 5c: (Patient 2) Transverse T1-fat saturated contrast-enhanced image at the same level as Figure 5b shows intensely enhancing mass with focal necrotic areas, infiltrating into the tibial cortex and medullary cavity.

image shows a circumferential mass with patchy intense enhancement and necrotic areas. The bone marrow of the tibia shows a focal moderately enhancing area beneath the mass.

Contrast enhanced transverse T1-fat saturated image demonstrates the longitudinal extent of the mass and infiltration into the bone.

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