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Case Report

# Radiological appearances of metastatic Marjolin ulcer in a chronic pressure sore: a case report and literature review ☆

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ABSTRACT

Marjolin ulcer is a type of aggressive ulcerating squamous cell skin tumor that typically develops in areas of previously traumatized, burned, chronically inflamed, or scarred skin. It typically occurs following a period of dormancy. We present a rare case of Marjolin ulceration with an unusual combination of continued non-compliance after diagnosis and 40 years of unusually long latency.



## Background

In developed nations, chronic pressure ulcers are a rare cause of Marjolin ulcer transformation. It is an infrequent and aggressive form of cutaneous cancer reported originating at sites of persistent non-healing wounds, pressure sores, venous stasis ulcers, lupus vulgaris, osteomyelitis, anal fistulae, pilonidal abscesses, and radiotherapy [1]. Although it is hypothesized that mutations in specialized cells due to chronic inflammation are the primary cause, there is no proven cause, criteria for the definition, categorization, pathogenesis, location, management, or prognosis of Marjolin ulcer [2]. Squamous cell carcinomas are the most common histological finding in Marjolin ulcer; however, malignant melanomas and basal cell carcinomas have also been documented [3]. Squamous cell carcinomas with Marjolin ulcers are more aggressive, have a higher risk of recurrence, metastasis rates, and high mortality than non-Marjolin ulcer Squamous cell carcinomas, and should be evaluated as a differential in any chronic wound to attain a better prognosis [4]. We present a rare instance of Marjolin ulceration with a unique combination of continuing noncompliance after diagnosis and 40 years of unusually extended latency.

☆ Competing Interests: The authors declare that they have no competing interests.

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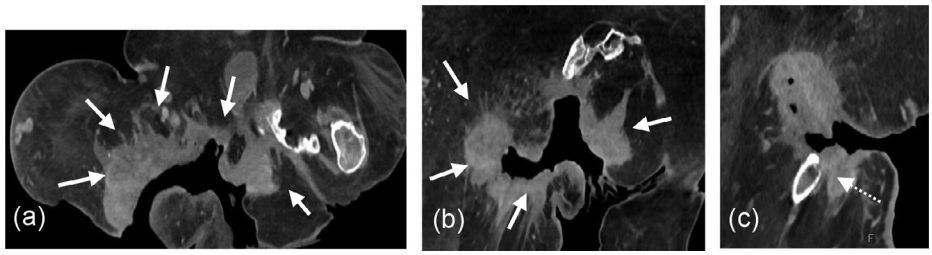


**Fig. 1 – CT images, axial (a), coronal (b), and sagittal (c), show a large circumferential ulcer at the right buttock with large width and markedly hypertrophied margins with spiculated edges (between the multiple white arrows in (a and b) and a deep extension eroding the posterior cortex of the proximal femur (white dotted arrow in c).**

## Case presentation

A 54-year-old woman was referred by the community nurse team for a Grade 4 pressure ulcer on her buttocks and sacrum. She was known to have Spina Bifida with congenital proximal femoral focal deficiency and was confined to a wheelchair for the previous 40 years. She had a chronic wound in the right buttock which required prior surgical interventions. There was a history of initial wound healing followed by recurrence. She continued to treat the wound by herself for the past 29 years and was not under the care of a wound nurse. A few months prior to her recent presentation, she experienced severe pain as well as noticed an increase in the size of the ulcer and contacted her community nurses for support. She was otherwise known to be healthy.

On inspection, the buttock and sacral wound measured approximately 17 cm in width × 8 cm in length × 9 cm in depth. It had one hundred percent granulation and rolled edges, accompanied by chronic blanching purple skin alterations in the surrounding area. The wound edges exhibited excessive granulation, were nodular, fragile, and quickly bled. No clinical indications of infection were observed, and there was no foul odor or sinus drainage. Given the clinical presentation and suspicion of osteomyelitis, magnetic resonance imaging (MRI) was sought.

Since the patient was initially unable to tolerate MRI, computed tomography (CT) was performed, which demonstrated a large circumferential pressure ulcer on the right buttock, with large width and deep extension. The ulcer was infiltrative with hypertrophied lobulated margins compared to previous old imaging. It was now also noted to extend up to the posterior cortex of the femur shaft, which showed long-standing chronic periosteitis and cortical sclerosis. Compared to previous imaging, the posterior femoral cortex exhibited new cortical erosions (Fig. 1). Additionally, few enlarged regional lymph nodes were noted.

Interpretation of MRI was challenging because of the altered anatomy. However, the ulcer edges were hypertrophied and of mass-like appearance, measuring about 4 cm in thickness, attaining low signal at T1WI and high signal at T2WI (Fig. 2). The cortical margins of the proximal femur showed bone edema confined to the eroded margins as described at CT images but there was no marrow replacement on T1WI and the edema was not extensive (Fig. 3).

The imaging findings were not conclusive of osteomyelitis, but concerns were raised regarding the unusual appearance of the ulcer's margins. Hence a specialist opinion and histological examination were recommended. The patient was seen by plastic surgeons who arranged for ulcer margin incisional biopsies.

Histology showed a well to moderately differentiated keratinizing squamous cell carcinoma invading the entire thickness of the specimens up to 10 mm. Within the desmoplastic stroma was a poorly differentiated, invading component with a diffusely infiltrative growth pattern. There was no perineural or vascular invasion found.

A subsequent staging CT scan of the thorax, abdomen, and pelvis revealed lung metastases (Fig. 4) and involvement of the inguinal and iliac lymph nodes.

Following a discussion with the patient, she opted for no further treatment and asked for a self-discharge from the hospital.

### *Diagnosis*

Metastatic Marjolin Ulcer

## Discussion

Marjolin ulcer is a relatively rare disease that affects approximately 0.05% of spinal cord injury patients with chronic pressure ulcers [5]. Tissue samples should be obtained from chronic pressure ulcers that have been present for more than 10 years to rule out cancer, especially if the ulcer's morphology changes, such as the development of exuberant granulation and/or bleeding [6]. The median age at diagnosis is in the fifth decade, ranging from 18 to 84 years, and men are 3 times more likely to be affected than women [7]. The time between the initial occurrence of an ulcer and the development of a Marjolin ulcer might range from 18 months to 30 years [8]. However, the latency period in our case was longer than anticipated due to inherited abnormalities and early-onset paraplegia. Malignant degeneration of chronic pressure ulcers and other chronic skin ulcerations may become more prevalent as the life expectancy of patients with spinal cord injuries increases [9]. As a result, careful inspection and regular, continuous monitoring are required for early diagnosis and treat-

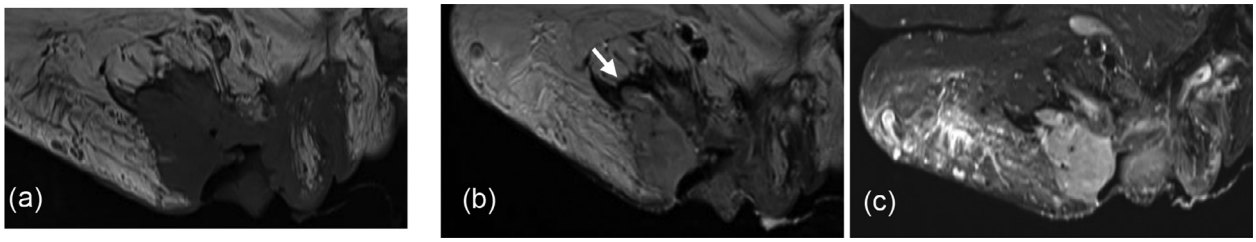


**Fig. 2 – Axial T1WI (a), T2WI (b), and STIR (c) MRI images show the marked soft tissue thickening at the ulcer margins with a mass-like appearance of a low signal at T1 (a) and high signal at T2 and STIR (b). The eroded cortex of the proximal femur is seen at the base of the ulcer (white arrow in b).**

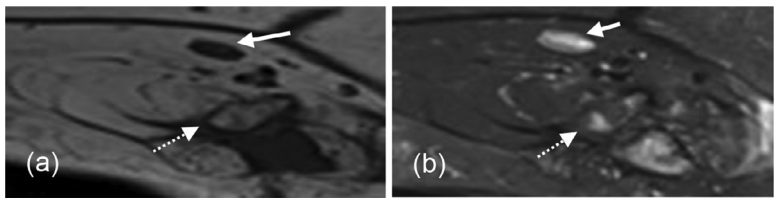


**Fig. 3 – Axial T1WI weighted (a) and STIR (b) MRI images at more caudal sections, show the ulcer adhering to the femur posterior cortex (dotted arrow) with mild subcortical edema at STIR yet the T1WI marrow signal is not replaced. Also, note enlarged regional lymph nodes (short white arrow).**

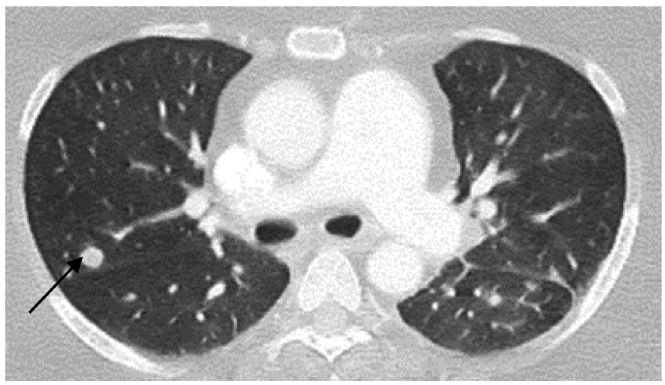

**Fig. 4 – Axial CT thoracic image showing right upper lobe metastatic pulmonary nodule (black arrow).**

ment. The most effective approach for minimizing malignant developments is to avoid pressure ulcers and surgically treat chronic ulcers [10].

Diagnosis of Marjolin's ulcer essentially depends on a low threshold of clinical suspicion and pathology of biopsy specimens. Imaging is valuable in evaluating the extent and depth of the ulcer, its relation to nearby structures and the extent of invasion if present. The presented case is unique in the unusual size and extent of soft tissue hypertrophy of the ulcer's margin upon which concern was raised. This reflects the patient's long noncompliance. Cross-sectional imaging, CT, and MRI are excellent to depict the amount of the tumor and the underlying bone destruction [11]. MRI has superior contrast resolution with the benefit of having T1WI and T2WI for optimal soft tissue characterization. Fat suppression sequences are superb for the detection of early and minor changes in the bone or soft tissues, providing high diagnostic sensitivity. T1-weighted images remain the best for differentiation of bone marrow edema from cellular infiltration as in infection and malignancy by the depiction of fat marrow replacement [12]. MRI with contrast enhances the delineation of the tumor's margin and depth as well as provides information on vital neurovascular structures [11,12]. The value of CT in the assessment of chronic pressure ulcers, especially deep grade 4 ulcers, cannot be overlooked. With its superior anatomical resolution images clearly depict the extent and pattern of periosteal reaction, amount of bone destruction and/or new bone formation, all of which are essential information for consideration by surgeons [17]. There is however some controversy in the literature regarding the reliability of bone changes in the diagnosis of complications in chronic pressure ulcers in particular osteomyelitis and malignancy. For instance, Smit et al determined in their study that periosteal reaction in chronic leg ulcers was nonspecific and did not aid in the diag-

nosis of Marjolin's ulcer. They however mentioned that large soft tissue mass was common in their patients and that malignant transformation was directly related to longer duration of ulcers [11]. Kolawale et al [13] described the periosteal reaction in tropical ulcers as the classic "ivory ulcer osteoma". A solid organized new periosteal bone in chronic leg ulcer is viewed as a benign bone response "ulcer osteoma", whereas lamellated periosteal reaction is suspicious of more serious complications like osteomyelitis [14–16].

Even with the most advanced diagnostic imaging, an expert radiologist can miss an initial diagnosis of malignant transformation of the ulcer. As new evidence is gained, close collaboration between the clinician and the radiologist should be maintained to re-evaluate the differential diagnosis especially when pressure ulcers have changed or become odd in appearance in a short time interval. We hope that this study may help to raise awareness among radiologists and clinicians to include this uncommon complication in their differential diagnosis in the proper clinical context.

There is currently no consensus on the duration and frequency of follow-up for patients with chronic pressure ulcers, and there are no guidelines for educating and monitoring high-risk patients. As such, early detection with a high index of suspicion and surveillance along with an early histological diagnosis are crucial for preventing and promptly treating cancer.

## Conclusion

Marjolin ulcer is a malignant transformation of chronic pressure ulcer, usually into aggressive metastasizing squamous cell carcinoma. The longer the duration of the ulcer, the higher the risk of malignant transformation. Surveillance and low index suspicion, as well as early pathological diagnosis, are crucial for appropriate treatment and a favorable prognosis. Cross-sectional CT and MRI are helpful in the depiction of the extent and depth of ulcer and relation to nearby structures, in particular bones. Hypertrophied soft tissue margins in a chronic ulcer should raise the suspicion of malignant change in the proper clinical context.

## Patient consent

Written informed consent for the case to be published (including images and case history) was obtained from the patient for publication of this case report, including accompanying images.

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