

CASE REPORT

Development of Marjolin's ulcer following successful surgical treatment of chronic sacral pressure sore

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Study design: Case report.

Objective: Report of an unusual case, where a Marjolin's ulcer that developed 2.5 years after surgical excision and successful closure.

Setting: Department of Plastic and Reconstructive Surgery and Burn Unit and the Clinic for Spinal Cord Injuries, Copenhagen University Hospital, Rigshospitalet, Copenhagen, Denmark.

Methods and Results: A 22-year-old man sustained a fracture with luxation of the 5th and 6th cervical vertebrae and loss of sensory and motor function after a diving accident (complete C8 lesion). During initial hospitalization, he developed a sacral ulcer, which more or less persisted for 38 years, despite several attempts of surgical and conservative treatment. At this time, the ulcer was finally excised and the wound closed successfully. Two and a half years later, however, the ulcer recurred. Biopsies showed squamous cell carcinoma and computed tomography, and magnetic resonance imaging scans revealed a soft tissue process over the sacral and coccygeal bones and massive destruction of these bony structures. The patient died 11 months later, despite surgical and radiation treatment.

Conclusion: The present case is unusual because, in spite of surgical excision and successful closure of the wound, the patient developed Marjolin's ulcer 2.5 years later. Yet it illustrates the primary importance of preventing the development pressure sores, of aggressive (surgical) therapy with healing when they do arise and of taking frequent biopsies in longstanding chronic ulcers to procure early diagnosis and treatment.

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Introduction

Malignant transformation is a rare^{1,2} but well-described complication of long-standing chronic pressure ulcers in spinal cord injury (SCI) patients.^{1–4} Carcinoma in chronic ulcers, known as Marjolin's ulcer, is also seen in connection with chronic ulcers of other etiologies, for example, in chronic burn scars, chronic fistulating osteomyelitis, venous stasis ulcers, traumatic wounds, anal fistula and urethrofistulae.^{1,2} The clinical course is usually rapid and fulminant with a high mortality rate.^{1–4}

Case Report

A 22-year-old man presented with an SCI after collision with a friend during a jump from a 3 m springboard. He sustained

a fracture luxation of the 5th and 6th cervical vertebrae and at discharge, he had a complete C₈ spinal cord lesion. In the initial stage, he developed a pressure ulcer over the left ischial area. In addition, he developed a large and a small pressure ulcer over the sacral and left trochanteric areas, respectively, which were treated conservatively. At final discharge, after 1 year and 9 months, there remained an ulcer, 1 cm in diameter, over the sacral area.

In the ensuing 20 years, the sacral pressure ulcer with a fistula persisted. At this time, the ulcer was excised and the defect closed with a fasciocutaneous Z-plasty. A fistula remained, but after revision healing was accomplished.

Twenty-five years after initial hospitalization, a pressure sore recurred over the sacrum. The patient declined to undergo surgical treatment.

Ten years later, the sacral pressure ulcer was treated at the local hospital with excision and closure, without success. A year later, renewed excision was made at department of plastic surgery, and the defect was covered successfully with a musculocutaneous transposition flap.

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Figure 1 Thirty-eight years after initial hospitalization, magnetic resonance imaging (MRI) showed an irregular soft tissue process, both solid and cystic in nature, over the sacral and coccygeal bones. Note the intact skin overlying the tumor.

Two and a half years later, the patient presented with increasing reflex sweating when pressure was applied to the sacral area. A computed tomography (CT) scan showed an exostosis with normally appearing overlying skin. Magnetic resonance imaging (MRI), showed an irregular soft tissue process, both solid and cystic in nature, over the sacral and coccygeal bones (Figure 1). A few days later, ulceration developed over the tumor. The patient was operated, but it was not possible to remove the tumor radically. Histological examination revealed well-differentiated squamous cell carcinoma. Radiation therapy was initiated, but afterwards, there was still histological verified carcinoma (Figure 2). A CT scan revealed extensive involvement of the sacral bone. The patient's condition gradually deteriorated and he died 40 1/2; years after his SCI and 11 months after the diagnosis of the carcinoma was made.

Discussion

Mustoe *et al.*¹ reported an incidence of squamous cell carcinoma of about 0.5% in 356 SCI patients. Eltorai *et al.*² found an incidence of Marjolin's ulcers in patients with SCI as low as 0.05%.

The causes of malignant degeneration are unknown but have partly been attributed to chronic irritation of the wound and to prolonged attempt at wound healing.⁴

Once carcinoma has developed, the survival rate is poor. For unknown reasons, especially in SCI patients 2 year mortality rates ranges between 67 and 80%.^{1,3} Most pressure sore carcinomas are situated in the sacral and ilial areas, which have a rich lymphatic drainage to the pelvic region explaining the high-metastatic rate. It has been speculated that tumor cells developing within scars and chronic wounds render tumor antigens inaccessible to the reticuloendothelial system. Thus, these cancers develop in an 'immunologically



Figure 2 Development of squamous cell carcinoma over the left sacral area, 2 1/2; years after successful surgical closure of a chronic pressure sore.

privileged site', leaving the body without an adequate cell-mediated response at the time of surgery.⁵

The patient in this case-report developed malignancy following closure of the wound. Procuring adequate tissue for histological evaluation during revision may therefore be of paramount importance to disclose early malignancy, as cure may be too late once there are clinical signs of malignancy.

The treatment of choice, in case of pressure sore carcinoma, is wide surgical with regional lymph node dissection. Radiation therapy has been used as palliation and the response to systemic chemotherapy is generally poor.²⁻⁴ This case demonstrates the necessity for rigorous and early surgical treatment with closure of chronic pressure sores to prevent malignant transformation. It is recommended that if a chronic pressure sore has been present for years, extensive biopsies should be taken to make sure that there has not already been malignant transformation that has not been suspected macroscopically. This may give a chance for earlier therapeutic intervention, although once the diagnosis of squamous cell carcinoma has been made, the long-term survival seems to be dubious.

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