



Marjolin's Ulcer Complicating A Pressure Sore

Ohnmar Htwe¹, Margaret Leow,² Amaramalar Selvi Naicker¹

¹Rehabilitation Unit, Department of Orthopaedic and Traumatology, University Kebangsaan Malaysia Medical Centre
²Plastic Surgery Unit, Department of Surgery, University Malaya Medical Centre

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ABSTRACT

Malignancy in a chronic pressure sore is rare among Marjolin's ulcers. Carcinomas arising in pressure sores are highly aggressive and usually fatal and therefore, they need to be treated more aggressively. We report a case of squamous carcinoma arising in a pressure sore of a 33-year-old paraplegic man who developed a sacral pressure sore with unstable healing for 10 years and a steady increase in size in the past 6 months was noted. The biopsy result showed a well differentiated invasive squamous cell carcinoma. A high index of suspicion is necessary for long standing pressure ulcer in this frequently neglected group of people to enable early diagnosis and treatment.

Key words: Marjolin's ulcer, squamous cell carcinoma, pressure sore

Basınc yarası ile komplike Marjolin's Ülseri

ÖZET

Marjolin's ülseri arasında bir kronik bası yarasında malignite nadirdir. Bası yaralarında gelişen karsinomlar oldukça agresiftir ve genellikle ölümcüldür, ve bu nedenle çok agresif şekilde tedavi edilmeleri gereklidir. Bu vakada 10 yıldır düzensiz iyileşme gösteren ve son altı ay içinde boyutu gittikçe artan sakral bası yarası gelişen 33 yaşında paraplejik erkek hastanın bası yarasında ortaya çıkan bir skuamoz hücreli karsinom olgusunu bildirdik. Biyopsi sonucu iyi diferansiye invazif skuamoz hücreli karsinomu gösterdi. Erken tanı ve tedavi için sıklıkla ihmal edilen bu hasta grubunda görülebilen uzun süreli bası ülserinde yüksek bir şüphe indeksi gereklidir.

Anahtar kelimeler: Marjolin's ülseri, skuamoz hücreli karsinom, bası yarası

Correspondence: Dr. Ohnmar Htwe
Rehabilitation Physician, Rehabilitation Unit, Department of Orthopaedics and Traumatology, University Kebangsaan Malaysia Medical Centre, Jalan Yaacob Latif, Bandar Tun Razak, Cheras (56000), Kuala Lumpur, West. Malaysia
Fax: 60391738609, HP: 60122074021
E-mail: htwe.om@gmail.com

INTRODUCTION

Marjolin's ulcer is a rare and an aggressive ulcerating cutaneous malignancy that arises on previously traumatized and chronically inflamed skin, especially after burns. It can be insidious and often leads to a poor prognosis. Deaths from Marjolin's ulcer are not uncommon. Meticulous wound care is a crucial step in prevention of these lesions (1). The majority of Marjolin's ulcers occur in wounds of the upper and lower extremities. Marjolin's ulcer appears to be preventable if early wound coverage is undertaken. Countries with limited access to medical treatment report a higher incidence of Marjolin's ulcers compared with more developed regions (2). A carcinoma resulting from a pressure sore is a very rare condition with an incidence of 0.5% (3).

CASE

We report a case of Marjolin's ulcer complicating a pressure sore. A 33-year-old man, who had been paraplegic for 13 years following a motor vehicle accident, had developed a sacral pressure sore with unstable healing for 10 years which necessitated repeated hospitalization for wound infection. The sore had been left open for years with epithelialization. The patient had the limited intact skin area due to right hip disarticulation and left above knee amputation. The wound had increased in size within 6 months prior to admission to our hospital in 2008 for non-healing infected pressure sore. Examination revealed a sacral ulcer measuring 20cm × 17 cm with foul smell discharge. The edges were undermined and filled with necrotic tissue. Although the patient had been prescribed broad spectrum antibiotics for infected wound, the infection was uncontrolled and continued to spread. The surrounding skin became hyper pigmented and developed nodules around the pressure sore within a week (Figure 1). The patient then was transferred to a Plastic Unit in another hospital for further management. A biopsy was taken from the sacral sore, showed a well differentiated invasive squamous cell carcinoma (Figure 2). CT scan of thorax, abdomen and pelvis showed multiple liver and lung metastases. Because of the patient's critical condition, by then no further surgery was done and the patient passed away 6 weeks later.

DISCUSSION

Cutaneous squamous cell carcinoma (SCC) is the second most common form of skin cancer and accounts for 20% of cutaneous malignancies (4). One of the possible risk factors for development of SCC is chronic inflammation which may lead to the development of squamous cell carcinoma irrespective of the underlying etiology. Both noninfectious inflammatory diseases and chronic infections have been associated with squamous cell carcinoma eg; Marjolin's ulcer. Burn scars are the best known chronic wounds to develop carcinomas. However the malignancy in chronic pressure sores is rare.

Celsus first noted, as early as the first century, the association between thermal burn scars and malignant degeneration. Later, a French surgeon, Jean Nicolas Marjolin, described a villous lesion that developed in degenerating scar tissue in 1828. Although others had previously described cancers developing in burn scar, Marjolin's name has been given to these malignancies (5). Mustoe et al reported squamous cell carcinoma arising in chronic pressure sores in paraplegic and tetraplegic patients (6). It is not common and consequently can easily be missed. It is said to have a worse prognosis than de novo SCC due to its high rate of metastasis (7).

Fleming et al. stated that over 90% of Marjolin's ulcers degenerate into malignancies of epidermoid origin, such as SCCs, basal cell carcinomas and malignant melanomas. Sarcomas can occur but are uncommon. Precise estimates of the true risk of malignant degeneration in a chronic wound or scar are difficult to obtain, as the data are predominantly derived from retrospective case studies (8). The latent transformation period of Marjolin's ulcers ranges between 25 to 40 years (9). Berkwits et al. reported squamous cell carcinoma in a pressure ulcer that had been present for 14 years (10). Tan et al reported a case of SCC in pressure sore with a short latency period of 6 months (11). Very chronic, simple ulcers are rarely tender or painful. In malignant disease there is both induration and pain which is of a hot, scalding, or darting character. Signs and symptoms associated with the development of the carcinoma include a change in the scar with formation of a mass or ulcer, possibly with an increase in pain, increasing discharge, foul odor and bleeding (12).

Pressure ulcer is a common complication in patient with spinal cord injury (SCI). Thirty three percent of patients had at least one pressure ulcer, of whom 13.6% had



Figure 1. Photograph showing sacral sore with rapid progression to surrounding skin a week after admission

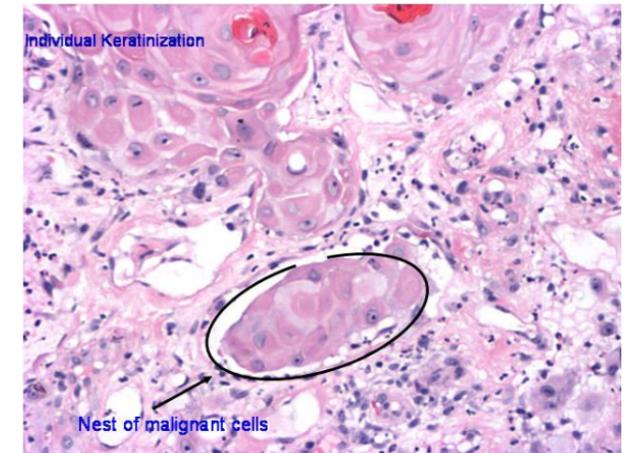


Figure 2. Photomicrograph of histo-pathological examination showing nest of malignant cells (Squamous Cell Carcinoma) - H&E staining (Magnification Power ×40)

stage 3 or 4 pressure ulcer according to a community-based survey of SCI patients (13). One of the possible causes of malignant degeneration of the wound is the persistent stimulation of marginal epithelium in non-healing wounds which in turn may lead to eventual loss of growth and neoplastic changes (1). These are rare tumours in which various aetiological factors have been incriminated, including repeated irritation, poor lymphatic regeneration, antibodies, mutations, and local toxins (14).

More recently, a theory of immunological isolation has been suggested, whereby lymphatic channel obliteration at the site of injury may decrease the delivery of antigen or specifically stimulated small lymphocytes to the regional lymph nodes from that site. This renders the site "immunologically privileged", allowing the development and progression of antigenically foreign tumour cells to go unchecked. Such cells may initially arise by spontaneous mutation or develop under the influence of viral or chemical carcinogens. Tumour antigen recognition may then be delayed long enough for tumours to reach "critical size", when immune mechanisms are no longer sufficient to prevent continued neoplastic progression (15). A rare atypical cellular occurrence in the wound healing process can also lead to malignancy. Endogenous growth promoting factors produced in the wound exudates which can act as co-carcinogens in genetically susceptible individuals (16).

Tissue biopsy is a definitive diagnostic tool and should include tissue specimens from both the centre and margins of suggestive lesions. Simple punch biopsy usually provides adequate tissue for diagnosis. A negative punch biopsy must be interpreted with caution because sampling error can occur (17). The most important diagnostic procedure for early diagnosis remains biopsy, which should be done on any lesion with recent changes. Dumurgier et al. advises biopsies of every pressure sore after the first decade (10).

Awareness of the malignant potential associated with chronic ulceration may allow early diagnosis and a decrease in the morbidity associated with advanced disease, such as radical node dissection or amputation. Although some aspects of treatment of Marjolin's ulcer remains controversial, several basic principles are necessary to practise in the treatment of long standing pressure sores. Preventive care is of greatest importance. In all wounds, infection should be treated early and adequate drainage should be provided when necessary. Infected wounds must be treated with appropriate antibiotics. In general, recurring ulcers should be excised even if they are not malignant and skin grafts or flaps should be used for coverage to facilitate complete healing as quickly as possible (18). There is a wide variety of suggested treatment protocols for this disease. A multitude of options and recommendations exists for the management of both primary lesions and regional

nodal metastasis. However, early diagnosis and radical excision are essential requirements.

Aggressive surgical therapy for pressure sore carcinoma is warranted because of the poor prognosis. Wide excision of the cancer is mandatory (19). Once a burn or skin trauma has occurred, care must be taken to ensure rapid and stable healing by skin graft or flap coverage. The involved skin lesions will not heal in the presence of tumor cells. Wide local excision with a margin of at least 1 cm of healthy tissue should be done in cases of Marjolin's ulcer. Amputation is indicated when wide local excision is prevented by deep invasion, bone or joint space involvement, infection or hemorrhage, or when excision would impair function and encumber the patient. Lifeso stated that wide local excision can be unreliable for grade II and grade III disease; therefore, amputation and prophylactic node irradiation is recommended (20). Long-term follow-up is recommended in all cases of Marjolin's ulcer.

In conclusion, pressure sore carcinomas appear to be a highly lethal condition. They are more aggressive with a higher mortality rate. If there is any suspicion, a biopsy should be performed. A high index of suspicion is necessary for long standing pressure ulcer in this frequently neglected group of people to enable early diagnosis and treatment. Early wound coverage is mandatory for non-healing ulcers.

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