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Case Report

Multiple Marjolin's ulcers arising from irradiated post-burn hypertrophic scars: A case report

Kevin J. Zuo^a, Edward E. Tredget^{b,*}^a Wound Healing Research Group, Division of Plastic and Reconstructive Surgery, Division of Critical Care Medicine, Firefighters' Burn Treatment Unit, Canada^b Department of Surgery, University of Alberta, 2D2.28 WMC, 8440-112 Street, Edmonton, Alberta T6G 2B7, Canada

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ABSTRACT ?

Marjolin's ulcer is an aggressive ulcerating cutaneous malignancy that may arise in chronically inflamed or traumatized skin. Frequently overlooked, this rare condition is classically associated with burn scars, with the process of malignant degeneration typically occurring over two to three decades. The most common histopathological pattern is squamous cell carcinoma; however, compared to typical squamous cell carcinomas, Marjolin's ulcers have an increased rate of metastasis. The correlation between radiotherapy for benign hypertrophic scarring and carcinogenesis is controversial, with few reports in the literature. We present a unique case of a 61 year old Caucasian male who was burned by scald at age 4, received radiotherapy for his post-burn hypertrophic scars, and later developed multiple Marjolin's ulcers on his left arm, chest, and right temporal scalp.

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1. Introduction

Marjolin's ulcer is an aggressive ulcerating cutaneous malignancy that may arise in chronically inflamed or traumatized skin. Ulcerating lesions in scar tissue were first described in 1828 by Jean-Nicholas Marjolin [1], and malignant degeneration of such wounds was recognized in 1903 by Da Costa [2]. Marjolin's ulcer is classically associated with burn wounds (76%), but may also arise from chronic wounds such as pressure sores (2.6%), venous stasis ulcers (6.3%), traumatic wounds (8.1%), and osteomyelitis (2.6%) [3]. The major risk factors for malignant degeneration in burn scars include healing by secondary intention, non-healing wounds, and fragile scars that are easily traumatized [4]. The mean patient age at diagnosis of Marjolin's ulcer is 52.1 years with an average latency period of 28.7 years following the initial injury

[3]. Squamous cell carcinoma (SCC) and basal cell carcinoma (BCC) are the most common histologies, at 71% and 12%, respectively. SCC occurs most commonly on the lower extremity (41%), while BCC most commonly affects the head, neck, and scalp (76%) [4]. As Marjolin's ulcers are very aggressive with a high rate of metastasis (35% for burn scar SCC versus 0.5–16% for standard SCC) [5], radical excision with possible radiotherapy is the current standard of care, although there is no agreement on prophylactic lymph node dissection [6].

The correlation between hypertrophic scar radiotherapy and carcinogenesis is controversial, with few reports in the literature. The authors present a unique case of a patient burned 57 years ago who received hypertrophic burn scar radiotherapy and developed multiple Marjolin's ulcers on his left arm, chest, and right temporal scalp. This is the first report in the literature of successive SCC, BCC, and basosquamous

* Corresponding author. Tel.: +1 780 407 6979; fax: +1 780 407 7394.

E-mail address: etredget@ualberta.ca (E.E. Tredget).

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(BSC) Marjolin's ulcers in multiple locations on a single patient.

2. Case presentation

A man was burned by scald in 1934 at 4 years of age on the left arm, chest, and right side of his face. He was treated in hospital for a prolonged period of time in Great Britain. His burn wounds healed by secondary intention after 9 months of dressing changes (no excision or skin grafts). He later developed hypertrophic scars over the burn regions, which were treated with brachytherapy and external beam radiation (radiation type and dose unknown).

Fifty-seven years later (1992), the man (age 61), a half pack per day smoker, presented with a 15 cm × 10 cm fungating tumor on the dorsal, radial, and volar aspects of his left forearm which had grown rapidly in the past 4–6 months (Fig. 1). Radial and ulnar pulses were normal and the patient had good hand function. Cultures grew diphtheroids,

Streptococcus viridans, *Pseudomonas putida*, beta hemolytic *Streptococcus*, anaerobic gram positive and negative bacilli, spirochaetes, and *Staphylococcus aureus*.

Based on the history and physical examination, the presumed diagnosis was burn scar SCC (Marjolin's ulcer), but initial wedge biopsy demonstrated malignant BCC. CT and MRI showed local invasion of the tumor into the flexor digitorum superficialis muscle and radial artery of the left forearm. There was sparing of the median and ulnar nerves and no clinical or radiologic evidence of regional or distant lymph node metastasis. The malignant tumor was surgically resected with free margins on frozen section, the radial artery was reconstructed with an autogenous vein graft, and the defect was closed with a split thickness skin graft (STSG) with excellent post-operative hand function (Fig. 1). Subsequent histopathology using permanent sections of the entire lesion showed that the tumor was in fact well-differentiated SCC, with all margins free of tumor. A regional lymph node resected from the antecubital fossa with the original specimen showed no evidence of metastasis.

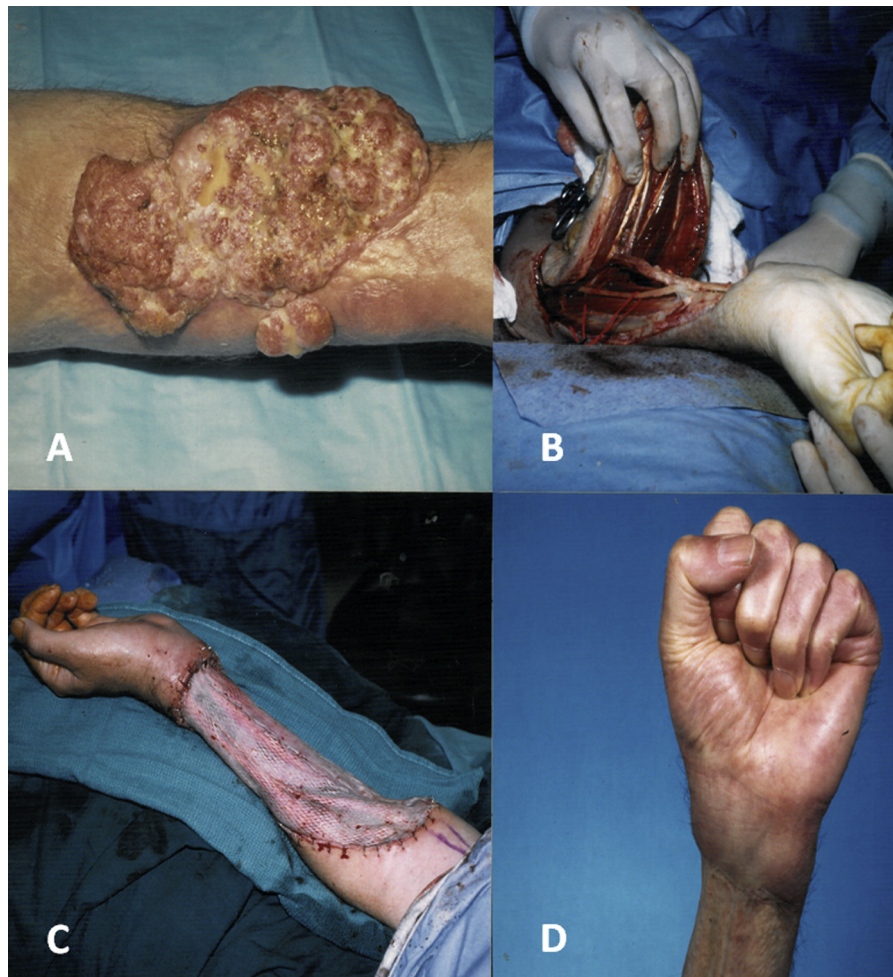


Fig. 1 – A squamous cell carcinoma of the upper extremity (A) 30 years after spontaneous healing of a thermal injury of the upper extremity and radiation therapy for the management of severe hypertrophic scarring. The tumor was removed at the level of the superficial flexor muscles of the forearm (B) and the arm was resurfaced with a split-thickness autograft (C), resulting in stable wound coverage with good hand function (D).

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