

Verrucous carcinoma: An unexpected finding arising from a burn scar



Claire E. Hannah, BS,^a Emily A. Weig, BS,^a Sigrid Collier, MD, MPH,^{b,c} Mary S. Stone, MD,^{d,e} Omar Juma, MD, MPH,^f Jane J. Mcharo, CO,^g Nkanyezi N. Ferguson, MD,^d and Karolyn A. Wanat, MD^h
Iowa City, Iowa; Minneapolis, Minnesota; Bagamoyo, Tanzania; and Milwaukee, Wisconsin

Key words: burn scar carcinoma; Marjolin ulcer; skin cancer; verrucous carcinoma.

INTRODUCTION

A malignant neoplasm arising in scar tissue is a well-known but rare phenomenon commonly called *Marjolin ulcer*, named after French physician Jean Nicolas Marjolin.¹ Marjolin's ulcer affects only 1% to 2% of thermal burn scars and most commonly takes the form of aggressive, poorly differentiated squamous cell carcinoma (SCC).² Verrucous carcinoma (VC), an uncommon, low-grade histologic variant of SCC, rarely develops in scar tissue, with only a handful of cases reported in the literature.³⁻⁷ Most of these cases involved middle-age men,³⁻⁶ with tumors developing an average of 2 decades after the inciting injury.^{3,4} There is only one report of a female patient with burn scar VC,⁷ and no known cases involving the upper extremity. We report an unusual case of VC on the proximal arm of a middle-age Tanzanian woman, arising only 3 years after severe thermal burn injury to the upper extremities.

CASE REPORT

A Tanzanian woman in her late 30s with epilepsy presented to a skin clinic in rural Tanzania, 25 km away from the nearest hospital, for evaluation of a 1-year history of an enlarging, painful growth on her left upper arm. Approximately 3 years prior, she sustained a severe scalding burn injury to her upper extremities from a pot of boiling water, resulting in extensive scarring. The growth started as a small lump within the scar tissue of the left arm approximately 2 years after the burn injury. Over the next year, the mass continued to grow in size and intermittently drained a foul-smelling substance.

Abbreviations used:

SCC: squamous cell carcinoma
VC: verrucous carcinoma

She received several courses of antibiotics from a local provider, which temporarily improved the exudate but had no effect on the mass. The tumor was interfering with her ability to care for her 4 children, including a newborn.

On physical examination, there was an approximately 9-cm exophytic, fungating, friable tumor on the lateral aspect of the left proximal arm, arising in extensive, mature scar. The tumor had malodorous, purulent exudate without surrounding erythema (Figs 1 and 2). Axillary, cervical, and submandibular lymph nodes were not palpable. She had no history of fever, weight loss, or any other changes to her health. Further workup was negative for hematologic abnormalities, and HIV testing was negative. Arm radiographs found absence of bone involvement. Because of financial constraints of the patient and lack of available diagnostic services in the area, no other imaging studies, laboratory work, or tissue cultures were obtained.

The patient was referred to a cancer treatment center in Dar es Salaam, Tanzania, 74 km from her home. However, she was unable to afford this option and instead elected for wide local excision under local anesthesia. One-centimeter excisional margins were selected out of concern for poorly differentiated SCC and inability to perform additional surgeries if surgical margins were positive. Histopathologic analysis of the

From the University of Iowa Carver College of Medicine^a; the Departments of Dermatology^b and Internal Medicine,^c University of Minnesota; Departments of Dermatology^d and Pathology,^e University of Iowa Hospitals and Clinics; Ifakara Health Institute, Bagamoyo^f; Bagamoyo District Hospital^g; and the Department of Dermatology,^h Medical College of Wisconsin.

Drs Hannah and Weig are co-first authors.

Funding sources: None.

Conflicts of interest: None disclosed.

Correspondence to: Karolyn A. Wanat, MD, 8701 W Watertown Plank Rd, Milwaukee, WI 53226. E-mail: kwanat@mcw.edu.

JAAD Case Reports 2019;5:225-7.

2352-5126

© 2019 by the American Academy of Dermatology, Inc. Published by Elsevier, Inc. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

<https://doi.org/10.1016/j.jidcr.2018.12.006>



Fig 1. VC arising in a burn scar. Clinical photograph shows an exophytic, fungating, friable tumor on the lateral aspect of the left proximal arm in a background of mature thermal burn scar.



Fig 2. VC arising in a burn scar. Close-up clinical photograph shows an exophytic, fungating, friable tumor on the lateral aspect of the left proximal arm in a background of mature thermal burn scar.

tumor found a hyperparakeratotic, verruciform, atypical epidermal squamous proliferation arising from the epidermis with a pushing border extending into the dermis (Fig 3). These findings were consistent with VC—an unexpected but favorable diagnosis. With daily dressing changes and a 15-day postoperative course of cephalexin, the surgical site was well healed by 3-week follow-up. The patient will continue to be clinically monitored for recurrence.

DISCUSSION

In 1828, Marjolin first described “thick woolly velvet” ulcers developing within a scar,¹ but it was not until years later that other clinicians identified the malignant nature of these ulcers and their association with previous thermal injury.² Marjolin ulcers most frequently affect the lower extremities of men in their fifth decade of life, developing an average of 30 years after the initial injury.⁸ Squamous cell carcinoma is identified in more than 70% of cases.² Basal cell carcinoma and melanoma are the next most common causes of Marjolin ulcer, but account for only 12% and 6% of cases, respectively.⁷ Verrucous carcinoma, an epithelioid variant of well-differentiated SCC, is rarely identified in a Marjolin ulcer, with very few cases reported.³⁻⁷

Classic VC presents as a nontender, papillomatous tumor with foul-smelling drainage, frequently occurring on the genitals (Buschke-Loewenstein tumor), palmoplantar surfaces (carcinoma cuniculatum), and

in the oropharynx (Ackerman tumor).⁹ It most commonly develops in middle-age, white men and is characterized by an indolent growth pattern and low risk of metastasis.⁹ Although the exact etiology is unclear, chronic inflammation may contribute to VC development, especially in cases in which the tumor develops outside of classical locations.^{3,5,9} Surgical excision is usually curative; risk of local recurrence is low and may be further reduced with Mohs surgery and adjuvant radiotherapy.^{3,9,10}

VC arising in a burn scar was first reported in 1981, when Aton and Kinstrey⁴ described a 60-year-old man who had VC within a childhood scar on the anterior shin. Since then, only a small number of cases have been reported.^{3,5-7} In several of these cases, the patient’s clinical course suggests that burn scar VC may behave differently and perhaps more aggressively than classic VC. Fazeli et al⁶ described burn scar VC arising on the foot of a man who was only 21 years old, and Huang et al⁷ described a lower extremity burn scar VC in a 60-year-old woman that recurred 2 years after full excision and skin grafting, necessitating above-the-knee amputation and lymph node dissection. Similarly, our young, female, HIV-negative African patient veers from the typical demographic of a patient with VC, and the tumor’s rapid development and short latency period further suggest a more clinically aggressive phenotype.

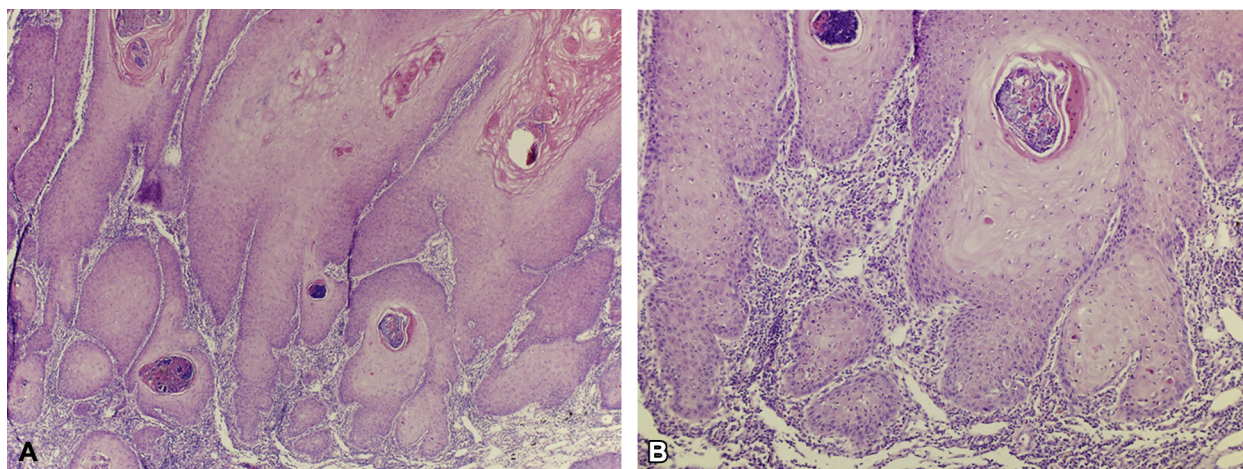


Fig 3. VC arising in a burn scar. Histopathology shows a verruciform, atypical epidermal squamous proliferation arising from the epidermis with a pushing border extending into the dermis. (Hematoxylin-eosin stain; original magnifications: **A**, $\times 4$; **B**, $\times 10$.)

We present this interesting and unusual case of verrucous carcinoma arising within scar tissue after a thermal burn sustained 3 years prior. Although a handful of cases of burn scar VC have been reported, this is the first case reported involving the upper extremity in a patient from a developing country. This rare diagnosis has important treatment and prognostic implications, especially in a resource-limited setting, and close clinical follow-up to monitor for recurrence is important.

REFERENCES

1. Marjolin JN. *Ulcere: Dictionnaire de Médecine*. Paris, France: Bechet; 1828.
2. Bazaliński D, Przybek-Mita J, Barańska B, Więch P. Marjolin's ulcer in chronic wounds — review of available literature. *Contemp Oncol*. 2017;21(3):197-202.
3. Diehl ES, Fleury RN, Ura S, Opromolla DV. Exuberant verrucous carcinoma arising from a burn scar. *Cutis*. 2007;79(2):133-135.
4. Aton JK, Kinstrey TE. Verrucous carcinoma arising from a burn scar. *Int J Dermatol*. 1981;20(5):359-361.
5. Brienza E, Albanese V, Armenise G, Miro A. Burn scars complicated by verrucous carcinoma — our experience. *Ann Burns Fire Disasters*. 1998;10(4).
6. Sadegh Fazeli M, Lebaschi AH, Hajirostam M, Keramati MR. Marjolin's ulcer: clinical and pathologic features of 83 cases and review of literature. *Med J Islam Repub Iran*. 2013;27(4):215-224.
7. Huang CY, Feng CH, Hsiao YC, Chuang SS, Yang JY. Burn scar carcinoma. *J Dermatolog Treat*. 2010;21(6):350-356.
8. Kowal-Vern A, Criswell BK. Burn scar neoplasms: a literature review and statistical analysis. *Burns*. 2005;31:403-413.
9. Koch H, Kowatsch E, Hödl S, et al. Verrucous carcinoma of the skin: long-term follow-up results following surgical therapy. *Dermatol Surg*. 2004;30(8):1124-1130.
10. Schwartz RA. Verrucous carcinoma of the skin and mucosa. *J Am Acad Dermatol*. 1995;32(1):1-21.