

# Giant-neglected facial Marjolin's ulcer associated with perioperative blood loss anemia

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## Access this article online

Website: [www.avicennajmed.com](http://www.avicennajmed.com)

DOI: 10.4103/ajm.ajm\_156\_19

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## ABSTRACT

Cutaneous squamous cell carcinoma (cSCC) is the second most common cancer in humans after basal cell carcinoma. Marjolin's ulcer is a form of cutaneous malignancy, and mostly represents cSCC, arising from chronic burns and wounds. We describe an interesting case of giant-neglected facial Marjolin's ulcer identified as cSCC associated with perioperative blood loss anemia. We also highlight its staging workup and the treatment options provided.

**Key words:** Anemia, blood loss, cell cancer, squamous, ulcer

## INTRODUCTION

Cutaneous squamous cell carcinoma (cSCC) is the second most common cancer in humans after basal cell carcinoma (BCC). Due to the shared lineage with epidermal keratinocytes, “keratinocyte carcinoma” is becoming the preferred term to refer to BCC and SCC instead of the traditional term “nonmelanoma skin cancer.”<sup>[1]</sup> As most primary keratinocyte carcinomas have low metastatic potential, most of the identified lesions are localized and minimal risk. Metastatic cSCC is uncommon and comprises approximately 4%–5% of the cases.<sup>[1]</sup> Only a small fraction (0.4%) may present with giant tumors with a diameter of  $\geq 5$  cm and these are considered high-risk tumors with high morbidity and mortality.<sup>[2]</sup> Traditionally, the “Marjolin's ulcer” term is used to refer to SCC arising from burns' scars; however, currently, it encompasses all malignant tumors that primarily occur in body surface ulcers that include squamous cell carcinoma, BCC, melanoma, sarcoma, and others.<sup>[3]</sup> Although perioperative anemia is a very common finding reported in head and neck SCCs (HNSCC),<sup>[4]</sup> it is not reported with facial cSCC. We describe an interesting case of giant-neglected facial Marjolin's ulcer identified as cSCC associated with perioperative blood loss anemia, its staging workup, and the treatment options provided.

## CASE REPORT

A 63-year-old Caucasian male presented with a 3-year history of ulcerating lesion involving the left cheek and parotid gland. Three years ago, he started to have slow-growing lesion arising from a nonhealing wound that occurred after a left cheek traumatic injury by a tree branch. The lesion was neglected until it started to increase rapidly in size over the last 5 months when he sought treatment. He reported associated left-sided hearing loss, weakness in eye closing, and mandibular weakness, all on the left side. He denied weight loss, trismus, or any lymphadenopathy.

The patient lived in California and had excessive sunlight exposure before he moved to Arkansas, where he received treatment. He has a past medical history of asthma, and cSCC of the right cheek required Mohs surgery. He reported no medications use or allergies. He denied tobacco, alcohol, or recreational drug use or any occupational chemical exposure. No history of immunocompromised status or

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**Cite this article as:** Kaako A, Malkan RM, Goff CP. Giant-neglected facial Marjolin's ulcer associated with perioperative blood loss anemia. *Avicenna J Med* 2020;10:128-31.

immunosuppressive therapy reported. The patient's mother died from colon cancer at age 46, and his brother had a history of melanoma.

On physical examination, there was an extensive bleeding fungating growth overlying the left cheek, extended to the left helix root and tragus [Figure 1A–C]. The lesion hung over the external auditory meatus, which was intact. The tympanic membrane was intact. There was no significant submandibular or cervical lymphadenopathy. He had pale conjunctivae. The remainder of his physical examination was unremarkable.

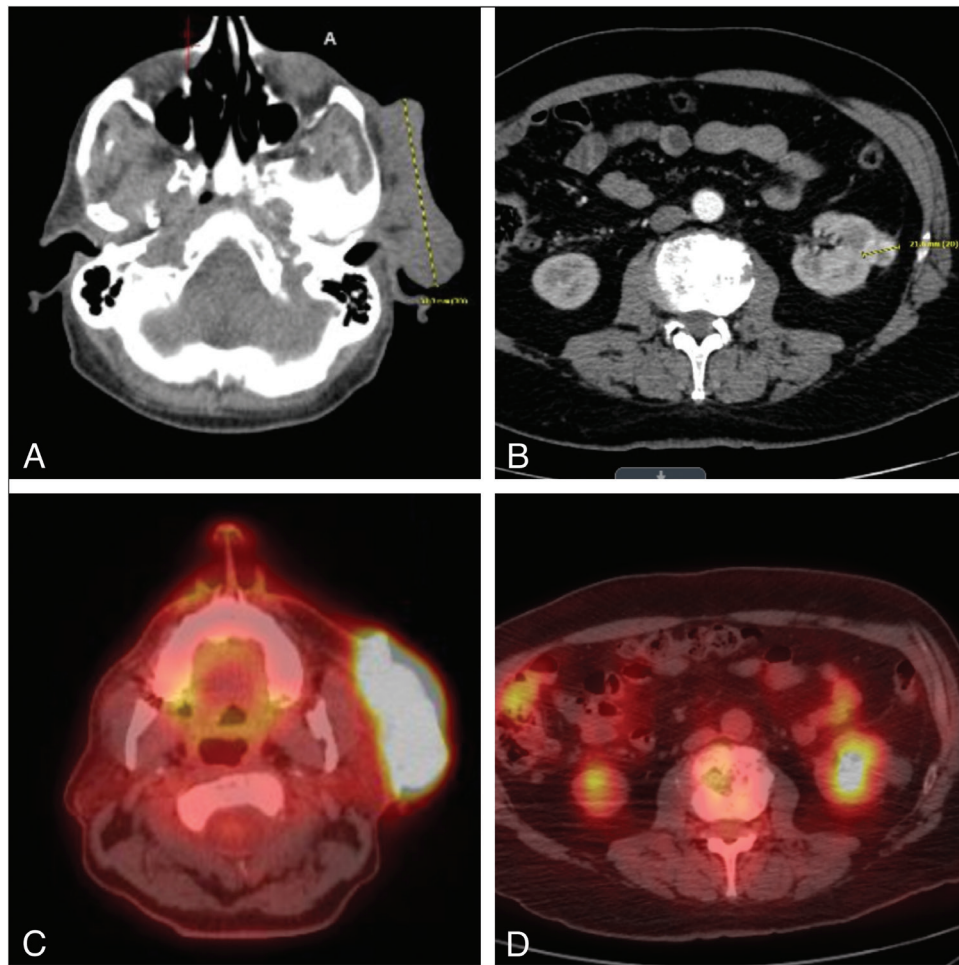
The patient was evaluated initially by a dermatologist, and his lesion biopsy showed poorly differentiated SCC. Given his extensive and high-risk SCC, he was referred to a head and neck surgeon. The computed tomography (CT) scan of the

head and neck showed an extracapsular invasion in the soft tissues of the muscles of mastication and the zygomatic arch [Figure 2A]. For the staging workup, the patient underwent a positron emission tomography/computed tomography (PET/CT) scan, which showed the giant invasive facial mass [Figure 2B] and interestingly showed an incidental finding of a 2.1 cm left cortical renal mass and bladder thickening [Figure 2B and D]. The patient reported no history of gross hematuria or flank pain. On the basis of his staging workup, he was considered to have a T3, N0, and MX poorly differentiated SCC.

The patient was scheduled for an elective surgical resection and a reconstruction flap. His preoperative evaluation revealed increased fatigue and dyspnea with exertion, palpitations, dizziness, and lightheadedness without syncope. His preoperative laboratory data showed



**Figure 1:** Giant-neglected facial Marjolin's ulcer. (A–B–C) Clinical presentation. (D) Defect after complex excision before flap application. (E) Intraoperative picture after the reconstruction of the left facial defect with an anterolateral thigh free flap with microvascular anastomosis. (F) Two days after surgery



**Figure 2:** (A–C) computed tomography (CT) and positron emission tomography (PET) scan of head and neck showing the giant invasive facial mass. (B–D) CT scan and PET scan of the abdomen and pelvis showing the incidental left renal mass

hemoglobin of 5.3 g/dL, mean corpuscular volume (MCV) 97.1, with normal B12, folic acid, and iron studies. He reported a long-term intermittent low-volume oozing from the facial lesion, which increased to an intermittent frank bleeding over the last 3 months. He denied hematochezia, hematemesis, hematuria, or coffee-ground emesis. Despite blood transfusion of two packed red blood cells (RBCs) units in the outpatient settings, his hemoglobin improved only to 6.5 g/dL. Due to his blood loss anemia, he was hospitalized preoperatively and received another unit of packed RBCs for medical optimization before his planned surgery.

He underwent an excision of the left zygomatic bone, left total parotidectomy with facial nerve sacrifice, and selective left neck dissection levels of I–IV [Figure 1D]. The reconstruction of the left facial defect was performed with an anterolateral thigh free flap with microvascular anastomosis [Figure 1E and F]. Pathology studies showed deeply invasive squamous cell carcinoma invading the parotid gland. All the margins and excised lymph nodes were negative for cancer.

Upon his postoperative follow-up, he was, expectedly, noted to have iatrogenic Bell's palsy. His ophthalmology examination revealed no corneal epithelial defect and he was started on high-viscosity artificial tears. The radiation oncology team recommended adjuvant radiation therapy to the deeper structures of the operative site and he is scheduled to undergo an intensity-modulated radiation therapy (IMRT) to preserve salivary function and minimize the risk of flap failure. For his renal mass, the patient was evaluated by urology and based on the peripheral location, renal cell carcinoma was suspected. He is scheduled for percutaneous CT-guided cryoablation after recovery from the facial reconstructive surgery.

## DISCUSSION

Marjolin's ulcers are form of cutaneous malignancy, and mostly represent cSCC, arising from chronic wounds. The malignant transformation rate was reported as 1%–2%.<sup>[2,3]</sup> Ultraviolet radiation exposure among fair-skinned individuals is the primary etiology

for keratinocyte carcinoma with higher incidence among individuals who have male gender, older age (>65 years), and lighter skin color,<sup>[1]</sup> which seems to fit the patient's presentation based on his demographics and location. Other risk factors include ionizing radiation, immunosuppression, chronic inflammation, arsenic exposure, human papillomavirus, and genetic disorders.<sup>[5]</sup> In his case, chronic inflammation occurred due to his neglected facial nonhealing wound. His presentation was consistent with high-risk cSCC,<sup>[6]</sup> which usually requires multidisciplinary approach. His management required collaboration among dermatologist, head and neck surgeon, plastic surgeon, internist, ophthalmologist, urologist, and radiation oncologist.

The lungs, liver, brain, and bone are the most frequent sites for distant metastases of cSCC.<sup>[7]</sup> Although it is not entirely excluded, the patient's kidney tumor was felt to be incidental finding and not directly related to his cSCC. Although anemia is very common in HNSCC and has prognostic implication,<sup>[4]</sup> symptomatic blood loss anemia has not been reported in cSCC.

As the population ages, keratinocyte carcinoma incidence is expected to increase dramatically over the next couple of decades. Focus on preventive measures and more public awareness of the carcinogenic effect of sunlight exposure is crucial to tackle the rising number of skin cancers.<sup>[1]</sup>

### Acknowledgement

We would like to acknowledge David G. Grant, MD, FRCS for his great contribution by providing excellent care for this patient and reviewing the manuscript.

### Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

### Financial support and sponsorship

Nil.

### Conflicts of interest

There are no conflicts of interest.

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