

Contents lists available at [ScienceDirect](https://www.sciencedirect.com)

International Journal of Surgery Case Reports

journal homepage: www.casereports.com

Marjolin's ulcer of the forearm from 30-year-neglect of external fixator

Haidar Kassir^a, Mohamad K. Moussa^{b,*}, Firass El Hajj^a, Walid Kheireddine^a,
Mohammad O. Boushnak^b^a *Departement of Orthopedic Surgery, Saint George Hadath Hospital, Beirut, Lebanon*^b *Orthopedic Surgery, Lebanese University, Faculty of Medical Sciences, Beirut, Lebanon*

ARTICLE INFO

Article history:

Received 19 January 2021

Received in revised form 28 January 2021

Accepted 29 January 2021

Available online 9 February 2021

Keywords:

Marjolin ulcer

Chronic infection

Neglected external fixators

ABSTRACT

BACKGROUND: Marjolin's ulcers are a rare form of malignancy that present at regions exposed to chronic infection. They present with a clinical triad of nodularity, induration, and ulceration greater than 3 months.**CASE REPORT:** We present herein, an extremely rare case of Marjolin's ulcer of the forearm, secondary to osteomyelitis, resulting from a 30-year neglect of external fixator used to treat a war injury of the forearm.**DISCUSSION:** Marjolin's ulcers are classically encountered in lower extremities at sites of burns, trauma or complicated wounds. In the upper extremity however, they are seldom mentioned in literature. The presence of risk factors raise the suspicion of the disease.**CONCLUSION:** Marjolin's ulcer is rare sequelae of chronic wound infection. Patients often present after a latency period with exacerbated pain, discharge, and exophytic mass. This disease should be suspected in every case of chronic ulcer, where histological studies of the lesion must be conducted to exclude or confirm the diagnosis.© 2021 The Author(s). Published by Elsevier Ltd on behalf of IJS Publishing Group Ltd. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

1. Introduction

The first description of Marjolin's ulcer was by Dr Jean-Nicolas Marjolin in the year 1828 [1]. The main cause of these lesions is chronic inflammatory reactions in wounds that failed to heal, where the time frame needed for malignant degeneration is several years [2,3].

Potential etiologies include burn eschars, pressure sores, venous stasis ulcers, traumatic wounds, osteomyelitis, and fistulas [4]. Patients usually present with symptoms including pain, bleeding, discharge, and growing exophytic masses that are refractory to treatment [5]. Once malignant transformation occurs, these lesions can invade deeper tissues and surrounding structures, requiring aggressive surgical treatment.

This report discusses a rare condition of marjolin's ulcer due to chronic infection on top of a 30-year neglected external fixator of the forearm. This case was reported in line with the SCARE criteria [6].

2. Case presentation

A 60-year-old male patient presented to the outpatient clinics with a complaint of left forearm pain, swelling, ulcerations, and purulent discharge. Upon inspection, the patient was found to have an external fixator of the left forearm. Detailed history was taken from the patient and his wife; the external fixator was put in place 30 years ago, following a gunshot injury during the Lebanese civil war, and the patient lost follow-up due to psychological issues. During that time, he had multiple pin tract infections that were treated with oral antibiotics throughout the years. 2 years ago, he sustained an injury to his left forearm upon falling down, and as a result, started having persistent pain, purulent discharge, and skin changes that looked like flourish nodules (Fig. 1). In spite of the above, he did not seek any medical advice until 1 month prior to presentation, after the smell of the discharge and the topography of his forearm compelled his wife to force him to seek medical care.

X-rays of the left forearm were conducted during the first visit, and they showed (Fig. 2) external fixation hardware of an old radius fracture with sclerotic changes of the mid and distal shaft of the radius, in addition to bony deformity of the distal radius, which might be correlated with chronic osteomyelitis. CT scan of the thorax, abdomen and pelvis was also carried out and showed no lymph node or distant metastasis. MRI of the forearm showed mal-limited ulcerations involving the skin and subcutaneous tis-

* Correspondence author.

E-mail addresses: Dr.haidarksr@gmail.com (H. Kassir),
mhamadmoussa71976798@gmail.com (M.K. Moussa),
firass.elhajj@hotmail.com (F. El Hajj), drwalidkh@yahoo.fr (W. Kheireddine),
mohammadboushnak@hotmail.com (M.O. Boushnak).



Fig. 1. Photograph of the left forearm of the patient showing flourish nodularity, purulent discharge and ulcerations with ill-defined borders, with the external fixator that was introduced 30 years ago. (A) Lateral side photography, (B) palmar side photography.

sues, spanning more than 17 cm in length and 6 cm in width, from the proximal third of the forearm all the way to the wrist and the hand. There were also signs of chronic osteomyelitis with a fistulous tract from the intramedullary canal of the radius, leading to a cutanea-subcutaneous collection proximally and medially forming an abscess 3 × 2 cm in dimensions (Fig. 3). Incisional biopsy was performed to confirm the suspected diagnosis, where histology showed an invasive, moderately differentiated (T4N0M0) squamous cell carcinoma consistent with Marjolin’s ulcer. Based on all collected data, the decision was made to proceed with above-elbow amputation.

Surgery was performed without complications, and follow-up was done on a weekly basis to check the wound. Finally, sutures were removed 21 days after the index procedure, and the wound appeared to heal quite well. Follow-up in the first 2 years post-

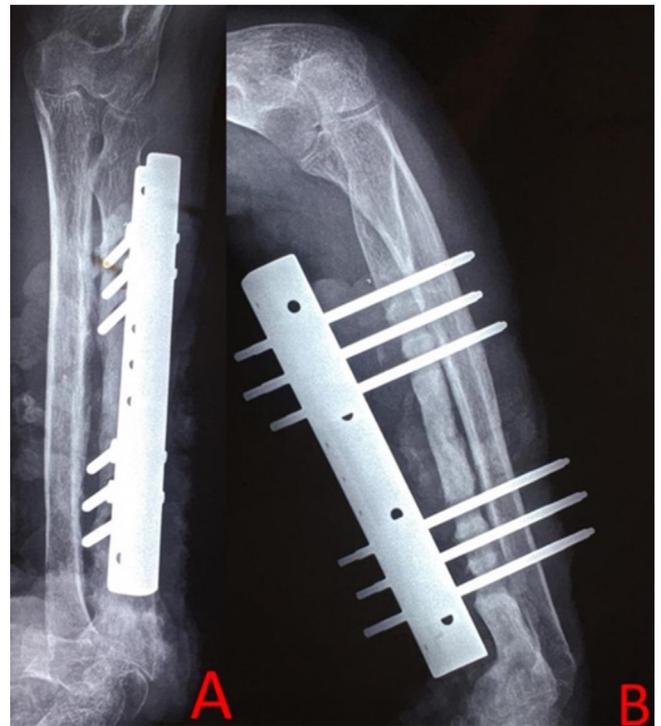


Fig. 2. AP (A) and lateral (B) radiographs of the left forearm showing an external fixator on top of an old radius fracture, bony sclerotic changes of the middle and distal third of the radial shaft. Bony deformity of the distal radius that may be related to chronic osteomyelitis.

operatively was done every 3 months with detailed history and physical examination, where both local and regional lymph nodes were checked cautiously for any suspicious findings. During the third year, the patient was examined every 6 months following the same aforementioned process. 3 years after amputation, the patient remained symptom-free, without any regional or local lymph node findings suspicious for recurrence. Instructions were given to the patient to maintain close follow-up, in order to detect any recurrence in an early stage.

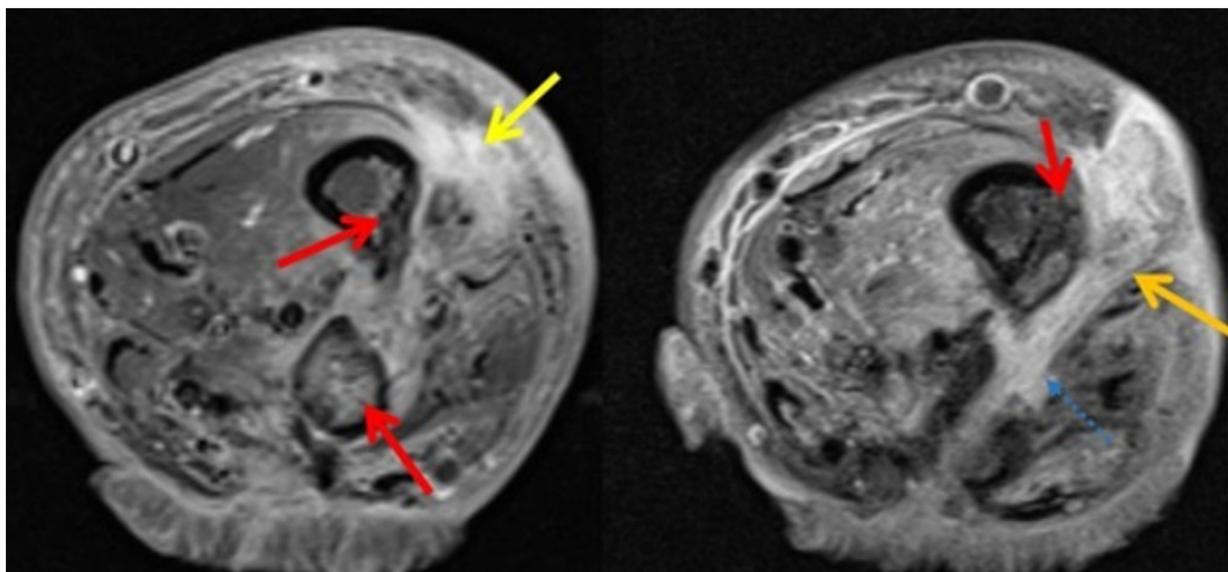


Fig. 3. coronal view MRI of the left forearm showing soft tissue collection (yellow arrow), osteomyelitis red arrow and fistulous tract (blue dashed arrow).

3. Discussion

After the first description of the French surgeon Jean Nicolas Marjolin in the year 1828, da Costa in 1923 used the term Marjolin's ulcer for the first time to describe malignant tumors occurring on top of burn injuries [1].

Marjolin's ulcer is a rare entity, estimated to occur in 1.7 % of all chronic wounds with a mean latency of 28.7 years [4]. A literature review done in the year 2009 showed that Marjolin's ulcers is caused mostly by burn wound with a rate of 76.5 % [7].

At present, Marjolin's ulcer denotes all neoplasms growing in scar tissues, chronic ulcers, and areas affected by inflammation [1]. Squamous cell carcinoma is the most frequent identified malignancy, although other types of malignancies have been described such as basal cell carcinoma [4]. After reviewing the literature, no cases of Marjolin's ulcer due to 30-year old neglected external fixator were identified.

The most common cause of Marjolin's ulcer is old burn scars, followed by malignant degeneration arising from chronic osteomyelitic fistula [2]. Chronic post-traumatic osteomyelitis and infected nonunions are complex problems that result in considerable morbidity and can thereby threaten the viability of the limb. The development of infection may result from compromised soft tissue and bone vascularity, systemic compromise of the host, and virulent or resistant organisms. Biofilm formation on implants and devascularized bone surfaces protects pathogens against host defenses and anti-biotherapy, and may lead to persistence of infection.

The reported latency period for the development of malignancy is between 11 and 75 years [3], with a mean of 30–35 years [4]. The younger the patient is at the time of injury, the more time it takes to undergo malignant transformation [8]. The clinical triad of Marjolin's ulcer consists of nodularity, induration, and ulceration occurring for a period greater than 3 months. Other signs and symptoms include rolled or everted wound margins [2], exuberant or excessive granulation tissue [7], foul smelling purulence, increase in size, bleeding on contact [9], crusting over [10], epithelial pearls [10], and pain [9]. Ulcerations associated with this entity often grow rapidly, with a flat surface and indurated elevated margins [10], but they may also be the slow-growing exophytic papillary type, which is less aggressive [11].

As a rule of thumb, biopsy should be performed on chronic non-healing ulcers, as it is the gold standard for diagnosing any malignant transformation [9]. There is positive correlation between the duration of ulceration and the risk of malignant transformation [10].

Although there is no definitive treatment protocol yet established for confirmed Marjolin's ulcer, therapy generally includes wide local excision with skin grafting [4], or amputation proximal to the lesion [2]. Refinements of the above procedures include free flaps, cryosurgery, and Mohs surgery [11], which includes complete circumferential peripheral and deep margin assessment using frozen section histology, whereby a surgeon serves as both surgeon and pathologist in the operating room; this is now considered to be the gold standard of treatments [10]. Other experimental treatments include carbon dioxide laser, intra-lesional interferon, and photodynamic therapy.

The 5-year cure rate is 90 % with Mohs surgery, compared to 76 % with surgical excision. Mohs surgery is however expensive and has a prolonged surgical time, and few doctors are adequately trained to perform the procedure [10]. Frozen section should be performed during the surgery, and if results showed positive margins, further resection or amputation is warranted [4].

Perhaps the most widely accepted treatment is amputation, although some recommend wide excision prior to amputation, if the latter would impair patient function [8]. Amputation is the

most definitive option for treating both the cancer and infection and is advised when either bone or joints are involved [4,9]. Ogawa et al. recommend amputation in grade II or III lesions, and wide local excision for very small or grade I lesions [12]. Finally, peri-operative management includes appropriate antibiotics following culture results and the removal of any foreign body [10].

Well differentiated lesions are less aggressive and therefore have better prognosis [10]. The 5-year survival rate is 40 %–69 % [4], 60 % for those with wide excision and 69 % for the amputation group [8]. After excision, the overall recurrence rate is 20 %–50 %, with 98 % of the ulcers recurring within 3 years [8]. Following amputation, the rate of metastasis is 20 %–35 % [12]. So long as the margins are clean following wide excision, there is no significant difference in recurrence between the latter and amputation [12]. The overall 3-year survival rate is 65 %–75 % [12], and the 10-year survival rate is 34 % [11]. However, for those with metastasis to the lymph nodes, the 3-year survival rate significantly drops to 35 %–50 % [9]. If patients survive past 3 years, they have good prognosis since 95 % of patients with metastasis present in the first 12 months [12].

4. Conclusion

Marjolin's ulcer is rare sequelae of chronic wound infection. Patients often present after a latency period with exacerbated pain, discharge, and exophytic mass. Treatment options vary according to local and regional factors. Early recognition and control of the disease allow for better control and relapse. This disease should be suspected in every case of chronic ulcer, where histological studies of the lesion must be conducted to exclude or confirm the diagnosis.

Patient perspective

The patient was satisfied with the result, and the patient returned to his regular activity 1 month post-operatively.

Declaration of Competing Interest

The authors report no declarations of interest.

Sources of funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors

Ethical approval

This type of study is exempt from ethical approval.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

Writing the paper: Haidar Kassir, Mohamad K MOUSSA, Mohammad Boushnak.

Data collection: Firass El Hajj, Walid Kheireddine.

Supervision: Haidar Kassir.

Registration of research studies

Not applicable.

Guarantor

Dr. Haidar Kassir.

Provenance and peer review

Not commissioned, externally peer-reviewed.

References

- [1] E. Copcu, Marjolin's ulcer: a preventable complication of burns? *Plast. Reconstr. Surg.* 124 (2009) 156–164, <http://dx.doi.org/10.1097/PRS.0b013e3181a8082e>.
- [2] M. Asuquo, G. Ugare, G. Ebughe, P. Jibril, Marjolin's ulcer: the importance of surgical management of chronic cutaneous ulcers, *Int. J. Dermatol.* 2 (46 Suppl) (2007) 29–32, <http://dx.doi.org/10.1111/j.1365-4632.2007.03382.x>.
- [3] E. Copcu, A. Aktas, N. Sişman, Y. Oztan, Thirty-one cases of Marjolin's ulcer, *Clin. Exp. Dermatol.* 28 (2003) 138–141, <http://dx.doi.org/10.1046/j.1365-2230.2003.01210.x>.
- [4] K.H. Chiang, A.S. Chou, Y.H. Hsu, et al., Marjolin's ulcer: MR appearance, *AJR Am. J. Roentgenol.* 186 (2006) 819–820, <http://dx.doi.org/10.2214/AJR.04.1921>.
- [5] M. Saaiq, B. Ashraf, Marjolin's ulcers in the post-burned lesions and scars, *World J. Clin. Cases* 2 (2014) 507–514, <http://dx.doi.org/10.12998/wjcc.v2.i10.507>.
- [6] R.A. Agha, T. Franchi, C. Sohrabi, G. Mathew, for the SCARE Group, The SCARE 2020 guideline: updating consensus Surgical CAse REport (SCARE) guidelines, *Int. J. Surg.* 84 (2020).
- [7] M.A. Kerr-Valentic, K. Samimi, B.H. Rohlen, J.P. Agarwal, W.B. Rockwell, Marjolin's ulcer: modern analysis of an ancient problem, *Plast. Reconstr. Surg.* 123 (2009) 184–191, <http://dx.doi.org/10.1097/PRS.0b013e3181904d86>.
- [8] B.B. Hill, D.A. Sloan, E.Y. Lee, P.C. McGrath, D.E. Kenady, Marjolin's ulcer of the foot caused by nonburn trauma, *South. Med. J.* 89 (1996) 707–710, <http://dx.doi.org/10.1097/00007611-199607000-00011>.
- [9] T. Bauer, T. David, F. Rimareix, A. Lortat-Jacob, Ulcère de Marjolin sur ostéite chronique: diagnostic et résultats du traitement: 7 cas [Marjolin's ulcer in chronic osteomyelitis: seven cases and a review of the literature], *Rev. Chir. Orthop. Reparatrice Appar. Mot.* 93 (2007) 63–71, [http://dx.doi.org/10.1016/s0035-1040\(07\)90205-6](http://dx.doi.org/10.1016/s0035-1040(07)90205-6).
- [10] B. Pekarek, S. Buck, L. Osher, A comprehensive review on Marjolin's ulcers: diagnosis and treatment, *J. Am. Col. Certif. Wound Spec.* 3 (2011) 60–64, <http://dx.doi.org/10.1016/j.jcws.2012.04.001>.
- [11] E. Aydoğdu, S. Yildirim, T. Aköz, Is surgery an effective and adequate treatment in advanced Marjolin's ulcer? *Burns* 31 (2005) 421–431, <http://dx.doi.org/10.1016/j.burns.2005.02.008>.
- [12] B. Ogawa, M. Chen, J. Margolis, F.J. Schiller, S.B. Schnall, Marjolin's ulcer arising at the elbow: a case report and literature review, *Hand (N. Y.)* 1 (2006) 89–93, <http://dx.doi.org/10.1007/s11552-006-9007-1>.

Open Access

This article is published Open Access at [sciencedirect.com](https://www.sciencedirect.com). It is distributed under the [IJSCR Supplemental terms and conditions](#), which permits unrestricted non commercial use, distribution, and reproduction in any medium, provided the original authors and source are credited.