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Marjolin's ulcer in a 20 years old split thickness skin graft on the knee—A case report

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ABSTRACT

OBJECTIVE: Marjolin's Ulcer (MU) is a rare cutaneous neoplasm arising in cikatrical tissue. Due to its typical clinical presentation as a non-healing lesion in scar tissue, the diagnosis can be delayed and even overlooked.
METHODS AND RESULTS: We present the case of an elderly woman who developed an ulcerated, exophytic lesion in a split thickness skin graft (STSG) on the lateral aspect of the left knee. Histology showed a radically excised highly differentiated squamous cell carcinoma (SCC) with keratine pearls and a component of basocellular carcinoma (BCC). The histological picture combined with the location and long time interval since the primary surgery made the diagnosis of MU highly likely.
DISCUSSION: Considering the risk of metastasis and mortality it is important to recognize the diagnosis and initiate adequate treatment.
CONCLUSION: The diagnosis of MU is clinical and confirmed by pathology. The typical long delay from the primary lesion to the malignant transformation might occlude the diagnosis. As such, a thorough anamnesis is essential in a non- healing ulcerated lesion in a cikatrical area to adequately diagnose and treat the condition.

1. Introduction

Split thickness skin grafts (STSG) are a reliable tool covering larger soft tissue defects. It does not, however, provide the restoration of adnexal tissues or original tissue architecture. As such, the function as a protective barrier is decreased, and subsequently also the resistance to mechanical damage and infections. The cikatrix can be hypo- or hyperpigmented, and the contraction can result in strictures, as are the features of cikatrical tissues. Marjolin's ulcer (MU) is a rare cutaneous neoplasm arising in scar tissue. The condition was first described by the French surgeon Jean Nicholas Marjolin in 1828 [1], as ulcerating lesions in cikatrical tissue. Even though he did not connect the condition to malignancies or squamous cell carcinoma (SCC), his name describes the condition where cikatrical tissues undergo malignant transformation. In this case we present a case of MU arising in a STSG 20 years after treatment for malignant melanoma. The work has been reported in line with the SCARE criteria [2].

2. Case

A 78 years old, otherwise healthy woman was referred to the department of plastic and reconstructive surgery for a non-healing wound on the lateral aspect of the left knee suspect of relapse of malignant melanoma. 20 years previously, the patient was treated for a thick malignant melanoma with a wide excision and covered with a STSG harvested from the anterior part of the left femur. There were no records available regarding the treatment, and histological records only stated, "malignant melanoma" removed with free margins without further description. During the past 4 months, she had noticed a nodular element laterally in the STSG, but attributed it to gardening. When the element persisted and intermittently bled and formed crustae, she contacted her general physician, who suspected infection and prescribed both topical and peroral antibiotics. When refractory to treatment, the patient was referred to a dermatologist, who suspected relapse of malignant melanoma and in turn referred the patient to our department. At the time of the referral, the lesion was freely mobile, measured 10 × 10 × 10 mm, and showed exophytic growth with an ulcerated surface. The surrounding tissues were without inflammatory reaction. There were no palpable regional lymph nodes. We decided to do a diagnostic excisional biopsy with a 5-mm margin. The procedure was performed in local anaesthesia, and we were able to close the defect by primary suture (Fig. 1). Histology showed a radically excised highly differentiated SCC with keratine pearls and a component of basocellular carcinoma (BCC), where the histological picture combined with the location and long time interval since the primary surgery made basis for the diagnosis of MU. The histology showed no signs of malignant melanoma or metastasis. The patient was offered wide excision to a total of 10 mm, and it was possible to close the defect without complications.

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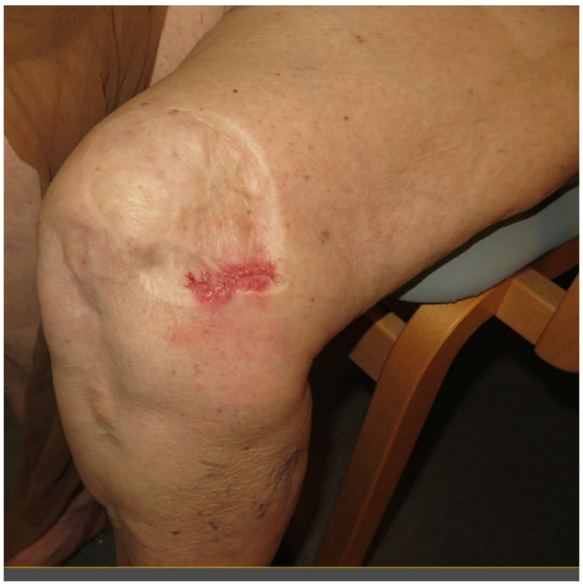

Fig. 1. STSG with cicatrix after 5 mm excision.

3. Discussion

MU is mostly seen in chronical cikatrical tissues, typically on the lower extremities (33%–53,3%) [3,4], mainly burns, osteomyelitic fistulas, in wounds caused by lupus and other chronical lesions. Its presence in skin grafts is to our knowledge scarcely described in literature. Non – healing wounds in cikatrical tissues, not responding to treatment should raise suspicion of malignancy. Biopsy, either interlesional or complete excision is important as this is the diagnostic tool. Some sources cite an incidence as high as 2% [3] of malignant transformation in chronical wounds, with a mean latency of 31 years [5]. The incidence is about 3 times as high in men as in women, and arises mainly in the fifth decade of life [6]. Histological analysis shows that most MU are highly differentiated SCC (71%), but both BCC (12%) and malignant melanomas (6%) are seen [3]. Subsequently the diagnosis is not only done on basis of the histological picture. The combination of histological analysis and an anamnesis with formation of a nodulus, induration and ulceration in cikatrical tissues dictates the diagnosis [6]. The exact mechanism of action is not clear, and many theories have been posed. Most likely it is a combination of several factors. Cikatrical tissues have reduced plasticity, and chronic irritations and shear forces such as in flexures and repeated traumas can result in atypia and constant mitotic activity. The constant repair and regeneration is postulated to trigger malignant transformation. In addition, cikatrical tissues often represent poorly vascularized tissues combined with impaired lymphatic drainage, and thus locally decreased immune response – and defence [3,4].

Literature recommendations for treatment vary from excision with or without sentinel node biopsy, radiation therapy, chemotherapy alone, or a combination [3,6,7]. There is currently no consensus or treatment protocol for MU in Denmark. The prognosis is dictated by size, lymph node status and metastasis at the time of diagnosis [6]. Kowal- Vern and Criswell report pathological regional lymph nodes in 22% of the cases, distant metastasis in 14% and relapse rates of 16% with an overall mortality rate of 21% in their case series of 412 patients [5]. In comparison, the rate of metastasis of an ordinary SCC is between 0,5% and 3% [8].

4. Conclusion

The diagnosis of MU is clinical and confirmed by pathology. The typical long delay from the primary lesion to the malignant transformation might occlude the diagnosis. As such, a thorough anamnesis is essential in a non- healing ulcerated lesion in a cikatrical area to adequately diagnose and treat the condition.

Conflicts of interest

None.

Funding

None.

Ethical approval

The institution (Zealand University Hospital) exempts the case report 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

(1) The conception and design of the study, or acquisition of data, or analysis and interpretation of data: Iselin Saltvig.

(2) Drafting the article or revising it critically for important intellectual content: Steen Henrik Matzen.

Registration of research studies

N/A.

Guarantor

Iselin Saltvig.

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