

Case Report

Marjolin's ulcer - dual lymph node metastasis with cystic degeneration: a rare presentation

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ABSTRACT

Marjolin's ulcer is a malignant tumour developing in chronic skin lesions (burn scar, non-healing wound etc.), with a mean time period of 36 years after sustaining burns. Majority of cases reported are squamous cell carcinoma, occurring in males and involving the extremities. Surgery remains the first treatment of choice (resection with 2 cm safety margin of healthy skin for primary squamous cell carcinoma in Marjolin's ulcers and 2.5 cm safety margin for recurrent cases). Recurrence after surgery and regional lymph node metastasis has been observed. We present a uncommon case in 55 year female with Marjolin's ulcer following burns scar over left abdominal region, who presented as dual regional, ipsilateral axillary and inguinal lymph node metastasis with cystic degeneration.

Keywords: Marjolin's ulcer, Squamous cell carcinoma, Lymph node metastasis

INTRODUCTION

Celsius first observed malignant transformation in burns scars in AD 100.¹ In 1828, French surgeon J N Marjolin² published an article on leg ulcers which showed warty changes. Term 'Marjolin's ulcer' was first used by Da Costa in 1903, for describing tumors arising from simple leg ulcers³. The term Marjolin's ulcer refers to malignant tumor developing in chronically inflamed or scarred skin lesions.⁴

The true incidence of Marjolin's ulcer is relatively unknown, with incidence ranging from 1.2 to 2%.⁵ Majority of cases reported are squamous cell carcinoma,⁶ over the extremities,⁷ commonest region of body involved are lower limbs 40%, 30% for head & neck regions, 20% for extremities & 10% for trunk.⁷

Though various unifactorial theories like Virchow's theory of chronic irritation,¹² Friedwald and Rouse co-carcinogenic theory,¹³ Treves and Pack's¹ toxin theory,

Ribet's theory of epithelial implantation⁹ etc. have been proposed for the pathogenesis of development of malignancy in scar patients, multifactorial theory have been propounded for malignant transformation, which proposes the combination of above theories as pathogenesis.

Two clinical types of Marjolin's ulcer have been described by Treves and Pack:¹ flat, indurated, infiltrative, ulcerative and exophytic, papillary forms.

Surgery remains the first treatment of choice, resection with 2 cm, safety margin of healthy skin for primary squamous cell carcinoma in Marjolin ulcers and 2.5 cm safety margin for recurrent cases¹⁴ is the preferred management followed by chemo/radiotherapy which is instituted on individual basis.

Lifeso and Bull describe histological classification with include grade I (well differentiated), grade II (moderately differentiated), grade III (poorly differentiated)

tumours.¹⁵ Staging, grading of the tumor, local recurrence and nodal metastasis are the main factors affecting the prognosis.¹⁶

CASE REPORT

A 55 year old female was presented to our surgical department with cystic lesions in left axilla and left inguinal region.

On examination, about single 5×5 cm cystic lesion was noted in left axilla and 4×4 cm cystic lesion in left inguinal region, having regular margins with restricted mobility.

Patient had a past history of burns injury, involving left side of chest and abdomen, which occurred when she was 6 years old for which she was conservatively managed.

About 2 years ago, at the age of 53 years, she noticed a non-healing, slow growing ulcerative lesion over the burns scar, but she ignored it. In June 2012, she reported to plastic surgery dept. for above complaint. On examination, ulcer was located in the left lateral side of abdominal wall, measuring about 15×10 cm, having everted margins and unhealthy granulation tissue. Edge biopsy was done from ulcer, which was reported as moderately differentiated squamous cell carcinoma (Marjolin's ulcer). No lymph node or regional metastases were noticed at that time. Subsequently, she underwent wide local excision with 2 cm margin followed by skin grafting.

Six months later, in January 2013, patient reported in surgery OPD with development of cystic lesions, noticed first in left axilla and then left inguinal region for which a thorough evaluation was done.

Routine investigations such as CBP, ESR, RBS, blood urea, serum creatinine and LFT's were within normal limits. She was non-reactive for HIV, HBsAg. Chest X ray was normal.

Ultrasound showed benign cystic space occupying lesion in left axilla and left inguinal region. MRI of left axilla revealed isointense solid component showing multiple thin septation suggestive of necrotic lymph node/cystic neoplasm/metastasis.

FNAC performed from both left axillary and inguinal cystic lesions, revealed few degenerated neutrophils, cyst macrophages in a proteinaceous background, suggestive of benign cystic lesion.

Patient was taken up for surgery for excision of cysts. Intraoperatively, axillary cyst contained about 50-60 ml of serosanguinous fluid. Cyst floor was firm to hard with friable necrotic material probably of nodal origin. Similar findings were noted in the inguinal swelling.

Biopsy was taken from both the swellings and sent for histopathology examination which revealed features of poorly differentiated squamous cell carcinoma deposits.

Subsequently she was referred to oncology institute for further management.



Figure 1: Inguinal region swelling.



Figure 2: Axillary region swelling.



Figure 3: Marjolin's ulcer - skin graft.

DISCUSSION

Majority of cases reported are squamous cell carcinoma,⁶ over the extremities,⁷ unlike in our case which involved the trunk region. They occur in every age group, most commonly in middle age,⁸ age of onset being 53-59 years; our case involved similar age group.

Latent period, that is time of interval between onsets of predisposing condition till the development of cancer is

supposed to be inversely proportional to the age of onset of predisposing condition.⁹ Shortest lag period ranged from 4 weeks¹⁰ to 70 years,⁶ mean time being 36 years after sustaining burns.

In our case, patient sustained burns at the age of 6 years and she developed non healing ulcer at the age of 53 years, i.e. after 47 years.

Based on latent period, Marjolin's ulcer have been classified as acute with lag period of <10 years and chronic ulcer with >1 year. Our case belongs to chronic Marjolin's ulcer category.

Most reports show male preponderance ratio of 2 to 3:1,¹¹ in our case, the patient being female. Commonest region of body involved are lower limbs 40%, 30% for head & neck regions, 20% for extremities and 10% for trunk.⁷

In our case, region involved was left side of abdomen i.e. trunk region, which is the least frequently affected site. According to classification of Marjolin's ulcer as described by Treves and Pack:¹ our case belonged to the ulcerative and exophytic forms.

It has been observed that malignant change begins at the edge of ulcer with base showing induration and necrotic material admixed with blood clots. These lesions spread early by direct tissue invasion and lymphatic invasion occurs in late stages, probably because of poor lymphatic drainage of scar tissue.

Lymph nodal metastasis has been considered as the most important prognostic factor. Metastasis to regional lymph node occurs in 2 to 6% cases. In our case we had dual lymphnode metastasis - ipsilateral inguinal and axillary nodes, without involving the contralateral side. This can be probably explained based on the anatomical location - involving the left lumbar/loin region, extending partly above and below the level of umbilicus, hence spreading to ipsilateral axillary and inguinal group of nodes.

Though it is said that, nodal metastasis are uncommon with Marjolin's ulcer, our case classically presented with dual nodal metastasis following 6 months of surgery for Marjolin's ulcer.

If HPE reveals an aggressive malignancy, post-surgery chemotherapy and radiotherapy should be considered apart from wide local excision to prevent recurrence as happened in our case. Marjolin's ulcer is a malignant squamous cell carcinoma occurring as post-burn complication and is more likely to metastasis.

Hence adequate surgical clearance with regular follow up may be helpful in preventing the recurrence of the disease, unlike in our case, where after doing the recommended surgery, it recurred as metastatic deposit.

This case is being presented for following reasons:

1. Squamous cell carcinoma developing as post burns complication after unusually long latent period of 47 years.
2. Uncommon dual lymph node metastasis, involving ipsilateral inguinal and axillary node without involving the contralateral side.
3. Cystic degeneration of the affected nodes.
4. Recurrence of tumor after recommended 2 cm of wide excision.
5. Region involved was left side of abdomen i.e. trunk region, which is the least frequently affected site.

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