

Clinical report

Marjolin's ulcers: the lasting effect of landmines in Cambodia

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Background: Marjolin's ulcer is a rare malignancy often associated with burn scars, traumatic wounds and chronic infections. Cambodia is still contaminated with landmines and unexploded ordinance, and a significant number of people suffer from related injuries, as well as latent development of Marjolin's ulcers.

Objectives: Report on the incidence of suspected and histopathologically confirmed cases of Marjolin's ulcers at the Children's Surgical Centre (CSC), Phnom Penh, Cambodia. Comment on 2 similar cases of landmine injuries to the lower limb resulting in Marjolin's ulcers.

Methods: Independent retrospective analysis of electronic and hardcopy patient records (January 2003 to January 2012) was performed by two examiners. Patients were selected based on primary and secondary inclusion criteria.

Results: Forty-eight patients were selected from the initial analysis and of those selected, 8 patients met secondary inclusion criteria. From these 8 cases, the preceding pathology included burns (62.5%), landmine blast injuries (25%) and chronic infection (12.5%).

Conclusion: The high prevalence of landmine blast victims in Cambodia suggests that there is likely to be a correspondingly high incidence of Marjolin's ulcers in the population. Improved surveillance for malignancy and prompt intervention in these patients may improve outcomes.

Keywords: Cambodia, landmine, Marjolin's ulcer, squamous cell carcinoma

"Marjolin's ulcer" is the term used to describe a rare malignancy arising from an ulceration that occurs in areas of cutaneous scars or chronic non-healing wounds [1, 2]. The French surgeon Jean-Nicholas Marjolin first described this phenomenon in 1828 in his article titled, "Ulcer" in "Dictionnaire de Médecine" [3]. Although the cancer is most commonly associated with burn injuries (approximately 75% of cases) [4], it has been reported in many other types of chronic injuries such as pressure sores, venous stasis ulcers, traumatic wounds, osteomyelitis, fistulas, leprosy ulcers, and lacerations [5].

Malignant transformation in chronic wounds tends to be slow, taking approximately 35 years on average [6, 7]. However, there have been reported cases of the development of these ulcers in a period of only a few months following the initial injury [8-10]. The major histopathologic type of Marjolin's ulcer is well-

differentiated squamous cell carcinoma [11] but basal cell carcinoma (BCC) has also been identified [12].

Here we report on the identification and treatment of Marjolin's ulcers in patients presenting to the Children's Surgical Centre (CSC), Kien Khleang National Rehabilitation Center, Phnom Penh, Cambodia. Due to the commonplace nature of landmine injuries in Cambodia, particular attention is paid to 2 cases of Marjolin's ulcer arising several decades post land mine injuries.

Method

To quantify the incidence of Marjolin's Ulcers at CSC, examiners A and B independently performed a retrospective analysis of patient records. Selected keywords (Marjolin, Squamous Cell Carcinoma, SCC, Basal Cell Carcinoma, BCC) were used to recruit an initial population of possible patients from both handwritten operation records and through the electronic patient record database; these keywords constituted the primary inclusion criteria. This initial analysis revealed 48 patients between January 2003 and January 2012.

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Subsequent analysis was performed by both examiners A and B: hardcopy patient records were reviewed for evidence of secondary inclusion criteria. These included evidence of a history suggestive of Marjolin's ulcer and at least one of the following; confirmed histopathology of well differentiated SCC or BCC, or a clinical diagnosis of malignancy. Following secondary analysis, 8 patients met these inclusion criteria.

Results

Retrospective analysis initially revealed 48 patients who met the primary inclusion criteria, of which 8 would meet the secondary inclusion criteria following subsequent analysis. Patient demographics (**Table 1**) revealed the average age of those presenting with Marjolin's ulcer to be 43.4 years. Of the 8 patients, malignancy arose from burns scars ($n = 5$; 62.5%), landmine injuries ($n = 2$; 25%) and sites of chronic infection ($n = 1$; 12.5%). Owing to the relative simplicity of the CSC electronic records system and the nature of hand written notes in the operation records, there is a high probability that whilst

performing the primary and secondary analysis, the examiners may have overlooked a number of patients with Marjolin's ulcers. This may suggest the incidence of Marjolin's ulcers at CSC is greater than uncovered in this review.

Case 1 (UNG-22985)

Fiftyfive-year-old male from Kampong Speu, Kong Pisei, presented to CSC on 4 October 2011 with a large fungating lesion on the anterior aspect of his right, proximal tibia (**Figure 1**). He had suffered a landmine injury to this area in May 1973. After radiological evaluation, a clinical diagnosis of SCC was made and on 25 October 2011 the patient underwent surgical resection of the tumour en bloc. This included removal of the surrounding skin and a portion of the proximal tibia below the tibial plateau, with preservation of the fibula. Following this, a free graft of the contralateral fibula was taken and used in conjunction with two internal fixation plates to bridge the bony defect (**Figure 2**). A soleus muscle flap was performed to close the wound.

Table 1. Demographics of eight patients presenting to CSC with Marjolin's ulcer between January 2003 and January 2012

Patient Number	Age	Gender	Address	Distance from CSC (km)	initial pathology
8137	35	Male	Kratie, Kracheh	252	Burn
9243	48	Female	Svay Rieng, Chantrea	162	Burn
18559	54	Male	Kampong Cham, Krouch Chhmar	177	Chronic Infection
18842	48	Female	Phnom Penh, Doun Penh	5	Burn
18885	61	Male	Kandal, Kien Svay	34	Landmine
19143	28	Male	PreahVihear, Kuleaen	28	Burn
19361	18	Male	PreahVihear, TbaengMeanchey	301	Burn
22985	55	Male	Kampong Speu, Kong Pisei	55	Landmine



Figure 1. Ulcer at presentation



Figure 2. X-ray post initial surgery

Infection at the surgical site and breakdown of the overlying skin resulted in further surgery (6 December 2011) to remove the internal fixation hardware and perform surgical debridement including removal of the necrotic fibula graft. External fixation was undertaken at this time.

A third surgery was performed on 17 January 2012 to fuse the tibiofibular joint with a bone graft harvested from the right iliac crest. External fixation with an Ilizarov apparatus was then applied as shown in **Figure 3**. At the time of writing this clinical report, the patient has made good postoperative progress and has been discharged.



Figure 3. X-ray post Ilizarov application

Case 2 (NGV-18885)

A 61-year-old male from Samraong Thum in the district of Kien Svay, Kandal (approximately 30 km southeast of Phnom Penh) presented to CSC with a 2-year history of a chronic ulceration on the postero-medial aspect of his left ankle (**Figure 4**).

This ulcer arose in the scar tissue from a landmine injury sustained 40 years earlier, in 1970. The wound measured approximately 12×10 cm. Radiological

imaging demonstrated the presence of shrapnel in the vicinity of the ulcer site (**Figure 5**).

The patient underwent a below knee amputation on 22 February 2010 to completely remove the lesion. Although the specimen was not sent for microscopic pathological evaluation, it was diagnosed as SCC on clinical grounds. Post-operative follow-up at two months revealed satisfactory wound healing. The patient was however, lost to follow up after this time.



Figure 4. Ulcer at presentation



Figure 5. Pre operative x-ray. Note the presence of shrapnel in the area of the ulcer

Discussion

The Children's Surgical Centre is a non-governmental organisation providing free rehabilitative and reconstructive orthopaedic, burn and plastic surgery, as well as ophthalmological services to Cambodian people. The CSC performs on average over 2280 rehabilitation consultations per year. Over a period of 9 years (2003 to 2012), 8 cases of Marjolin's ulcer were diagnosed and treated at CSC. The demographics of these patients are presented in **Table 1**. Not surprisingly, in the majority of cases (62.5%) the malignancy occurred in burn scars, the most common cause of this pathology [4]. Interestingly, there were two cases (discussed above) that showed the development of Marjolin's ulcer in areas affected by previous landmine injuries.

Cambodia is a country that was for many years torn apart by civil conflict and war, and still has much of its land contaminated with unexploded ordnance (UXO). Rural areas are particularly vulnerable due to the high cost of explosive removal campaigns, poor infrastructure and the population's overall unawareness of risk [13]. The injuries caused by such ordinances are varied, ranging from minor trauma to loss of limbs, disfigurement, blindness and death. The psychosocial impact of these injuries on victims and their families is immeasurable and often lifelong. In addition, the increased risk of developing Marjolin's ulcer within these wounds is just one more consequence of the ongoing collateral civilian involvement in the legacy of conflicts long since past.

In contrast to squamous cell carcinoma arising *de novo* in otherwise normal tissue, patients with Marjolin's ulcers suffer a worse prognosis. The 5-year-survival of typical SCC, compared with Marjolin's ulcer, is 85% versus 52% respectively and the 10-year-survival is similarly disparate, at 60% versus 34% respectively [4]. This discrepancy is potentially magnified in the developing world as resources aimed at early diagnosis, treatment and follow-up are limited. Although 75% of patients had their diagnosis confirmed by biopsy and histopathology, no patients underwent imaging studies to rule-out lymph node spread (exceeds 22% to 25%) or evidence of distal dissemination (6% to 14%) [4]. The majority of patients treated were also lost to follow-up in the short term and those that did return for outpatient review received no opportunity for long-term surveillance.

Conclusion

The large number of blast victims in Cambodia, in addition to the many other devastating effects, are at an increased risk of development of Marjolin's ulcers. The ongoing slow progress in removing this explosive material, combined with the often long delay between initial injury and development of malignancy, suggests that there are potentially many more cases of Marjolin's ulcers yet to develop, or yet to be identified, in the population. As such, special attention should be taken by medical personnel in the care of blast patients and a high index of suspicion of malignancy should be maintained in those with chronic ulcers. For those with landmine injuries (and burn injuries), long-term surveillance of wound sites by primary care physicians may be recommended. This could be invaluable in reducing the incidence of patients with Marjolin's ulcers presenting at an advanced stage, with a less favorable prognosis, and would help in some way to lessen the detrimental consequences that so many of these survivors are condemned to suffer for decades to come.

The authors have no conflicts of interest to declare.

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