

## Case Report

# Sympathetic dysfunction dermatitis in a revascularised upper extremity after near-total amputation – A case report and review of literature

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## ABSTRACT

Commonly reported complications after revascularisation or replantation of extremity are vascular thrombosis leading to complete or partial failure, bony non-union or malunion, joint stiffness and incomplete or abnormal sensory recovery. Sympathetic dysfunction dermatitis is an unreported complication after revascularisation or replantation surgery which results due to denervation of the extremity. We report a case of a young adult who developed eczematous dermatitis over the revascularised upper limb and discuss the role of sympathetic dysfunction in the development of these skin lesions. The patient was successfully treated with a short course of oral and topical steroids. Sympathetic dysfunction dermatitis is a rare form of skin eruptions occurring in the revascularised or replanted part of an extremity due to abnormal sympathetic function in the affected part.

## KEY WORDS

Eczematous dermatitis; replantation; revascularisation; skin lesions; sympathetic dysfunction

## INTRODUCTION

Frequently reported complications after replantation and revascularisation of upper extremity are vascular thrombosis leading to complete or partial failure, non-union or malunion of bones, joint stiffness and incomplete or abnormal neurological recovery including paraesthesia, dysaesthesia and hypoesthesia.<sup>[1]</sup>

Although abnormal skin eruptions and inflammatory skin lesions have been previously mentioned in patients with limb trauma and complex regional pain syndrome (CRPS),<sup>[2-6]</sup> the development of skin lesions and abnormal sudomotor function following replantation or revascularisation of the limb in such injuries has not been reported. We present the case of a young adult who

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developed eczematous dermatitis over the revascularised limb and discuss the role of sympathetic dysfunction in the development of skin lesions.

## CASE REPORT

A 24-year-old right-handed male reported to our emergency department with the history of trauma to his left forearm and hand while working on a wood-cutting machine. There was no associated injuries or comorbidities. There was near-total amputation of the left upper limb at two levels: at the junction of middle and lower one-third of forearm and at the level of metacarpal shafts. All the forearm structures were transected except preserved skin tag of 2 cm width on the ulnar side of the forearm [Figure 1]. The left thumb was also amputated at the level of mid distal phalynx. After the bony fixation, revascularisation of the limb was undertaken by repair of ulnar and radial arteries. Venous flow was established by repair of cephalic and basilic veins. Perineural repair of the ulnar nerve, the median nerve, superficial branch of radial nerve, and the lateral cutaneous nerve of forearm was carried out in an end-to-end fashion. Similarly, the first, second and third common digital arteries and nerves were repaired after the fixation of metacarpals. Thumb stump was closed primarily. Injury to reperfusion time was 7 h and total operating time was 13 h.

In the post-operative period, the patient developed vascular insufficiency of the distal part of the hand which could not be salvaged. We performed trans-metacarpal amputation of the left hand and covered the stump with a pedicled groin flap. Small raw area on the dorsum of the forearm was skin grafted. Complete healing of the wounds occurred after 1 month, at which time the patient was discharged from the hospital [Figure 2]. The patient was

referred to the physiotherapy unit for rehabilitation and was followed up in our outpatient department monthly for sensory and motor evaluation.

Four months after the discharge, the patient reported with acute-onset skin lesions over the revascularised part of the forearm and hand. The physical examination revealed weeping and crusting exudative lesions suggestive of eczematous dermatitis. The revascularised part of an extremity developed irregular erythematous plaques covered with yellowish thick crust. There were multiple erosions at places along with serous discharge. These lesions were strictly limited to the revascularised part of the limb; rest of the limb was normal [Figure 3]. The patient did not have any allergy and other reactive skin lesions in the past. Negative Patch test with standard series ruled out contact dermatitis. Skin biopsy revealed the findings of acute eczematous dermatitis.

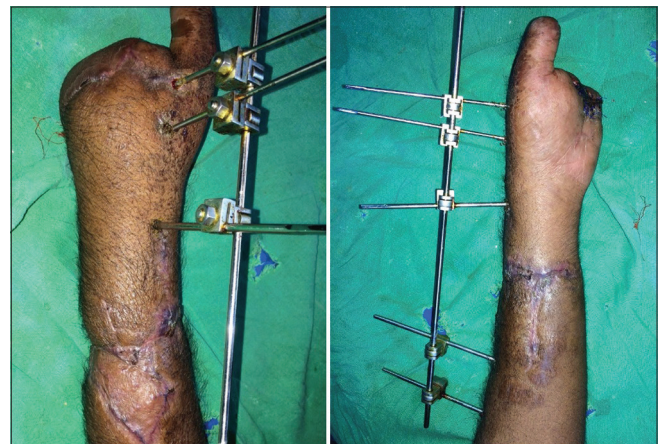
Several studies in the past have reported the association between autonomic dysfunction and skin lesions like atopic dermatitis, psoriasis and CRPS.<sup>[2-4]</sup> As the patient had suffered traumatic neuropathy, investigations were done to rule out autonomic dysfunction in the revascularised part. We performed Ninhydrin sweat test and Sympathetic Skin Response (SSR) test. Negative Ninhydrin sweat test indicated disturbance of sweating function.

SSR test was performed using a four-channel electromyography machine as described by Cicek *et al.*<sup>[2]</sup> The affected hand did not show any waveform indicating sudomotor dysfunction.

Sympathetic dysfunction in the revascularised part of the limb was confirmed by above investigations.



**Figure 1:** Pre-operative photograph and plain radiographs of the patient showing near-total amputation of left forearm and hand



**Figure 2:** Post-operative photographs showing complete healing after 4 weeks. Healed skin graft on the dorsum of forearm and healed groin flap at the trans-metacarpal amputation stump are seen



**Figure 3:** Photograph showing erythematous, exudating and crusting lesions of eczematous dermatitis 3 months postoperatively



**Figure 4:** All the lesions had completely healed after a short course of oral and topical steroids

Sympathetic dysfunction led to abnormal sudomotor and vasomotor functions and eczematous dermatitis in the revascularised part of the limb. Therefore, we coined the term *sympathetic dysfunction dermatitis* to describe the skin lesions developing in the revascularised or replanted part of an extremity.

The patient responded well to a topical cream of 2% fusidic acid and 0.1% betamethasone along with oral prednisolone for 1 week. All the skin lesions subsided with appearance of a normal skin texture [Figure 4]. However, there was one episode of recurrence 3 weeks after the resolution of the skin lesions which was treated as before. There was no further episode of dermatitis. The patient is presently on emollients to prevent recurrence.

To evaluate the sensory recovery, Semmes–Weinstein monofilament test and 2-point discrimination test (2PD) were performed at the 4<sup>th</sup>, 5<sup>th</sup>, 6<sup>th</sup> and 8<sup>th</sup> months postoperatively. On evaluation at the 6<sup>th</sup> month, the patient was free of skin lesions and his sensory recovery improved.

## DISCUSSION

Dermatitis at a surgical site was first reported by Carr and Rau in 1981<sup>[5]</sup> and later on by Bart in 1983<sup>[6]</sup> following vein graft harvest for cardiac surgery. The aetiology of this remained largely unknown until the mid-nineties when injury to the neural plexuses and localised venous congestion were implicated for dermatitis in these patients.<sup>[4]</sup>

A Japanese study cited saphenous neuropathy as the cause for occurrence of skin lesions. Injury to the nerve plexus leads to sympathetic dystrophy leading to skin

atrophy and a state of hyper- or hypo-hidrosis.<sup>[7]</sup> Local venous congestion after trauma can also lead to oedema of the skin, a pigmented purpura-like inflammatory dermatitis, and a non-immune bullous eruption.<sup>[7,8]</sup>

The association between autonomic dysfunction and development of skin lesions was emphasised by some studies. These studies reported the presence of sympathetic dysfunction in patients with atopic dermatitis, psoriasis and CRPS.<sup>[2-4]</sup> The unmyelinated C fibres are responsible for sudomotor activity which cause activation of sweat glands. Involvement of these fibres leads to dryness and itchiness of the skin, making it susceptible for the development of dermatitis.<sup>[2]</sup> Several interacting pathways are involved in causing skin inflammation, with autonomic and sensory nerves modulating the response.<sup>[9]</sup> Loss of modulatory effect of autonomic and sensory nerves after amputation may lead to skin inflammation in such patients.

More evidence into the relationship between sudomotor and sympathetic dysfunction and development of inflammatory skin lesions can be found in patients with hand surgery who develop CRPS.<sup>[10]</sup> CRPS is characterised by abnormal pain and sudomotor, vasomotor and trophic changes. After traumatic denervation, there is distal degeneration of small-diameter axons subserving nociception and sympathetic function.<sup>[11]</sup> There are reports of vascular and trophic changes along with increased interleukin-6 (IL-6), tumor necrosis factor- $\alpha$  and mast cell marker tryptase after trauma to the limb.<sup>[12,13]</sup>

It can be safely assumed that similar derangements take place in patients with traumatic amputations undergoing replantation or revascularisation. Along with sensory and



motor deficiencies, denervation of the distal part of the limb leads to trophic, vasomotor and sudomotor changes which lead to the increased likelihood of developing dermatitis in the amputated part.

Hruza and Hruza<sup>[14]</sup> found that this type of dermatitis typically occurs between 1 and 9 months and is associated with traumatic neurosensory deficits. Dermatitis improves with the improvement in sensory deficit.<sup>[13]</sup> This finding was observed in our patient where dermatitis was active between the 4<sup>th</sup> and 5<sup>th</sup> post-operative months and thereafter it subsided with the improved sensory recovery of the affected limb. There may be variable clinical presentations. Cutaneous manifestations can be seen in the form of cellulitis, dermatitis or xerosis.<sup>[8]</sup>

Zakrzewska-Pniewska and Jedras observed decreased sweating, hypohidrosis and skin xerosis as markers of autonomic dysfunction and that autonomic dysfunction can be successfully diagnosed with SSR test.<sup>[15]</sup> SSR test can be effectively used to evaluate the presence or return of autonomic function in patients with replantation or revascularisation. It can be a useful addendum to nerve conduction study and electromyography in monitoring return of neurological function.

Dermatitis due to sympathetic dysfunction after trauma responds well to treatment with short course of oral and topical steroids. However, there is a chance of recurrence of dermatitis until adequate sensory recovery has occurred.<sup>[7,8]</sup>

After replantation or revascularisation of the extremity, the primary focus remains on the survival of the limb and its functional rehabilitation. Skin changes might receive less attention and subsequently remain unreported. This report of sympathetic dysfunction dermatitis as a new complication after revascularisation or replantation of limb which should be kept in mind. Further studies are required to assess the prevalence and incidence of this entity.

## CONCLUSION

Sympathetic dysfunction dermatitis is a rare form of dermatitis seen in patients with traumatic denervation of the limb. It should be regarded as a potential complication of denervation of the extremity, which is expected to subside after reinnervation. Appropriate measures to keep the distal replanted part well hydrated may prevent this complication. Sympathetic dysfunction dermatitis responds well to oral and topical steroids.

## Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given his consent for his images and other clinical information to be reported in the journal. The patient understands that name and initial will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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## Conflicts of interest

None.

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