

Post Burn Cutaneous -Lymphangioma, Verrucous Type. A Case Report

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The cavernous lymphangiomas are frequently encountered as superficial lesions predominantly seen in skin and mucous membranes. They may sometime follow surgery or radiotherapy, they rarely develop in scars after a burn injury or in a congenital lymphadematous extremity without prior surgery or radiation. Here, we describe a case of an unusual superficial cutaneous cavernous lymphangioma of verrucous type in a 26 years old female patient following burns, clinically simulating squamous cell carcinoma.

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The cavernous type of lymphangioma, which constitute the majority of lymphangiomas are usually superficial and occurs predominantly within the skin or mucous membranes. These tumours have predilection for head and neck region, where they may extend deep into the muscles. They may also rarely involve the deep structures of the upper arm, mediastinum, retroperitoneum, abdominal cavity etc¹. Lymphangiomas secondary to trauma, such as surgery or burns are infrequently documented. Further verrucous forms are extremely uncommon and we report a case in a young Bahraini female.

THE CASE

Twenty six years young Bahraini female presented with the history of multiple papillomatous lesions on the left thigh region for over a year. The lesions were pale white, nodular, warty in appearance, and of varying in dimensions. Healed scars of the burns were present surrounding the lesions. There was a history of deep burns in this region during her childhood period. The patient did not give any history of loss of appetite or weight. Systemic examination revealed no abnormality. Haemogram and blood chemistry were within normal limits. There was no regional lymphadenopathy or abdominal organomegaly. These lesions had recurred twice consequent to excision and this was the third recurrence.

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Pathological Findings

Grossly multiple skin lesions having warty, nodular patterns were noticed on the surface measuring from 1.5 x 0.5cm to 2.5 x 1.0cm dimensions. The cut sections showed pale brown area in the stroma. Microscopic examination from different sites revealed similar morphological features in all the samples studied. The soft tissue lesion was covered by hyperkeratotic hyperplastic stratified squamous epithelium in verrucous pattern at places (Fig 1). The characteristic feature is the presence of large cystic dilated lymphatic channels lined by flattened endothelium predominantly in the subepidermal region and superficial dermis closely applied to the papillary dermis (Fig 2). Some of the channels contained pale eosinophilic fluid material and few red cells. Few vessel walls contained scant smooth muscle blending with the fibrous stroma. The intervening stroma was made up of fibrous tissue.

Figure 1. Dilated lymphatic channels covered by hyperkeratotic hyperplastic stratified squamous epithelium in verrucous pattern. H&E x 400

Figure 2. Subepidermis and superficial dermis with large cystically dilated lymphatic channels lined by flattened endothelium. H & E x 100

DISCUSSION

In the past, the word hemangioma was the generic term commonly employed for any kind of vascular lesions. In 1982, Mulliken and Glowacki¹ introduced a new classification based on the clinical and histological characteristics of the lesion, namely hemangioma and vascular malformation. In order to select the form of treatment and

timing, Jackson et al² reclassified the vascular anomalies according to the vascular dynamics of the lesion, ie. low flow or high flow. This classification is depending on the speed of flow through the lesion and the rate of shunting between the arterial and venous components. Based on these criteria, the current classification is as follows:

1. Hemangioma
2. Vascular malformations
 - a. Low flow lesion (venous malformation)
 - b. High flow lesion (arteriovenous malformation)
3. Lymphatic malformation (lymphovenous malformation)

Most lymphangiomas represent malformations rather than true neoplasms and are thought to result from failure of the lymphatic system to communicate with the venous system. It may even be acquired due to burns, radiotherapy etc. Although the complications of burns are many, the frequently encountered are keloid or hypertrophic scar, Marjolin's ulcer, non-healing ulcer etc. The lesions, grossly looking like keloid or hypertrophic scar and having dilated vascular channels in the underlying tissue, the so called cavernous type lymphangiomas are well described³. Three cutaneous cavernous lymphangiomas have been reported which occurred after surgery or radiotherapy for breast carcinoma and one in the scar after a burn injury in the series of lymphangiomas documented by Lars-Gunnar Kindblom and Lennart Angervall³. Single cases of superficial cutaneous lymphangioma (lymphangioma circumscriptum) have also been recorded following radical mastectomy and radiotherapy^{4,5} and spontaneously in a congenital lymphadematous extremity not subjected to surgery or irradiation⁶. However verrucous forms clinically resembling squamous carcinoma is rare.

Grossly, these have a spongy appearance and are multiple with brownish red papules. Characteristically, the covering squamous epithelium shows acanthosis and hyperkeratosis, as well as some areas of thinning, others present with hypertrophy. Unusually, the lesions may have a verrucous appearance. On light microscopy, cavernous lymphangiomas are composed of communicating open lymph spaces that run in different directions and have highly variable shapes, either empty or filled with lymph; they may also contain lymphocytes and occasionally, erythrocytes. Lymph thrombus and calcifications may occur³. Subcutis and deeper lymphangiomas, are frequently composed of vessels having a well organised mature smooth muscle component, which may be abundant and blend with the fibrous stroma. The vessels are lined by one or more layers of flattened endothelium, sometimes assuming a cuboidal or epithelioid appearance. Characteristically, there is a well developed fibrous stromal component around the vessels, which may blend with an abundance of adipose tissue in a lobular fashion specially occurring in the subcutis region. Verrucous cutaneous cavernous lymphangiomas should be considered in cases of unusual lesions developing consequent to lacerations, trauma or radiotherapy in the long term.

CONCLUSION

This case illustrates verrucous lymphangioma, a rare complication developing in 26 years old female following burns.

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