Ann Dermatol Vol. 31, Suppl, 2019
pISSN 1013-9087 · eISSN 2005-3894
https://doi.org/10.5021/ad.2019.31.S.S20

BRIEF REPORT

Unilateral Cutaneous Vasculitis on Lower Limb in Patient with Unilateral Lymphedema

Jong Sic Kim, Yu Ri Woo, Miri Kim, Hyun Jeong Park

Department of Dermatology, Yeouido St. Mary's Hospital, College of Medicine, The Catholic University of Korea, Seoul, Korea

Dear Editor:

Leukocytoclastic vasculitis (LCV) is a disease which is histologically characterized by leukocyte infiltration and rupture around the vessels, and also exhibits damaged vascular walls. The common causes are bacterial infection, intake of medications, malignancy or idiopathic diseases[1]. It is usually found clinically on the natal area of a recumbent patient or both lower limbs and it means that the hydrostatic pressure or stasis may play an essential role in the pathogenesis of the disease[1,2].

A 77-year-old female patient presented with painful, unilaterally involved, purpuric papules on the left leg that had appeared 2 days previously (Fig. 1A). We received the patient's consent form about publishing all photographic materials. The lesion first appeared on the left ankle and spread rapidly toward the proximal side of the leg, while the opposite leg was not involved. There was no history of infection or medication other than warfarin that may have contributed to induction of the skin lesion. The patient was regularly consuming oral warfarin to control lymphedema of the left leg that occurred after surgery and radiation therapy for cervical cancer 20 years ago. Laboratory findings showed mild elevation of liver enzymes, while other figures indicating infection were within the normal range. Evidence of deep vein thrombosis or tumor embolization was not found in the Doppler ultrasound examination. Findings from skin biopsy of a purpuric papule were consistent with LCV (Fig. 1B). Based on those

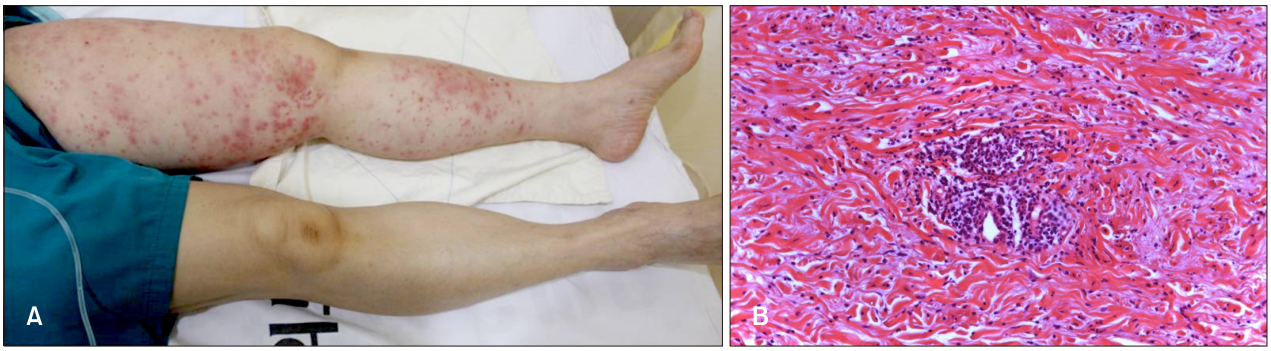

Fig. 1. (A) Unilaterally involved purpuric papules on the left lower leg. (B) Histopathologic examination revealed the fibrinoid necrosis of vessel wall and perivascular neutrophilic infiltration with leukocytoclasia (H&E, ×200).

Received July 18, 2018, Revised September 29, 2018, Accepted for publication November 1, 2018

Corresponding author: Hyun Jeong Park, Department of Dermatology, Yeouido St. Mary's Hospital, College of Medicine, The Catholic University of Korea, 10 63-ro, Yeongdeungpo-gu, Seoul 07345, Korea. Tel: 82-2-3779-1230, Fax: 82-2-783-7604, E-mail: hjpark@catholic.ac.kr
ORCID: https://orcid.org/0000-0002-0138-9885

findings the patient was diagnosed as LCV associated with lymphedema, and was treated with oral methylprednisolone (0.5 mg/kg/d) for 1 week and then progressive resolution of the rash was observed.

This case presentation was considered a rare case as unilateral LCV was diagnosed through skin biopsy of a patient who displayed unilateral lymphedema on the lower limb. Searches of the literature, revealed only 3 previous cases of unilateral LCV like this patient[3-5]. The underlying disease reported in the previous literature were diabetes and hypertention[3], chronic renal failure and paroxysmal atrial fibrillation[4], ankylosing spondylitis and familial Mediterranean fever[5]. Hence, no conditions were found to be associated with unilateral hydrostatic pressure. The authors of these reports considered the bed-ridden state of the patient[4], the chronic deep vein thrombosis[5] and miscellaneous vascular malformation[3] to be the cause of the disease. In this case, the patient had suffered from unilateral lymphedema for the past 20 years, hence, it is possible the increased hydrostatic pressure may have accounted for the pathogenesis of the immune-mediated purpuric lesions found in the LCV. Disruption of the lymphatic system could conceivably result in stasis with circulating immune complexes and cutaneous inflammation. Although oral warfarin could also contribute to develop LCV, drug-induced LCV usually occurs symmetrically. Thus, for this case, LCV-associated with unilateral lymphedema could be a better explanation.

In conclusion, the authors hereby present this rare case of cutaneous vasculitis with unilateral location following chronic lymphedema. Clinicians should be aware of the fact that chronic lymphedema is associated with the development of LCV, and therefore need to appreciate that LCV could be clinically distributed on the affected side of lymphedema.

ACKNOWLEDGMENT

This research was supported by the Basic Science Research program and Creative Materials Discovery Program through the National Research Foundation of Korea (NRF), which is funded by the Ministry of Education, Science and Technology and the Ministry of Science, ICT and Future Planning (2015R1C1A2A01055073, 2016M3D1A1021387).

CONFLICTS OF INTEREST

The authors have nothing to disclose.

ORCID

Jong Sic Kim, https://orcid.org/0000-0002-4091-390X
Yu Ri Woo, https://orcid.org/0000-0003-2903-9534
Miri Kim, https://orcid.org/0000-0001-5167-3449
Hyun Jeong Park, https://orcid.org/0000-0002-0138-9885

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