CLINICAL IMAGE

Fever and generalized erythema caused by subcutaneous panniculitis-like T-cell lymphoma

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A 72-year-old woman presented with generalized erythema, gradually increasing for 2 months, and fever—for 1 week. The erythema first appeared on the lower limbs and expanded to the trunk and upper limbs. The patient had a history of cholecystectomy for gallstones and left mastectomy for breast cancer. A physical examination revealed erythema and edema all over the body, including the trunk and particularly on the extensor and flexor aspects of the upper and lower limbs, respectively (FIGURE 1A–C). Additionally, recesses corresponding to the lesser and greater saphenous veins were observed on the flexor aspects of both lower limbs, suggesting subcutaneous inflammation (like the groove sign; FIGURE 1C). Thus, subcutaneous panniculitis was considered the cause of fever and erythema.

Blood tests revealed increased levels of lactate dehydrogenase (399 IU/l) and soluble interleukin-2 receptor (3563 U/ml). Contrast-enhanced computed tomography of the chest, abdomen, pelvis, and lower limbs revealed diffuse, enhancing, infiltrative lesions under the skin over the entire body (FIGURE 1DE). Skin biopsy specimens obtained from the lower limb flexor aspect, including subcutaneous tissue, revealed a mild lymphocytic infiltration

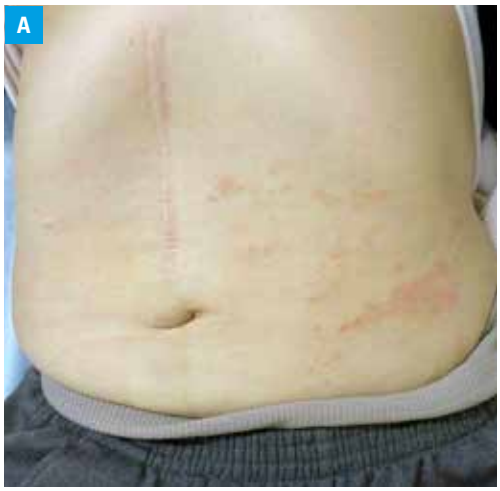

FIGURE 1 A, B, C – erythematous lesions all over the body and recesses corresponding to the lesser saphenous vein on the flexor aspect of the right lower limb; D, E – contrast-enhanced computed tomography of the abdomen and lower limbs showing diffuse, enhancing, infiltrative subcutaneous lesions
(go to page 868 for panels C, D, E)

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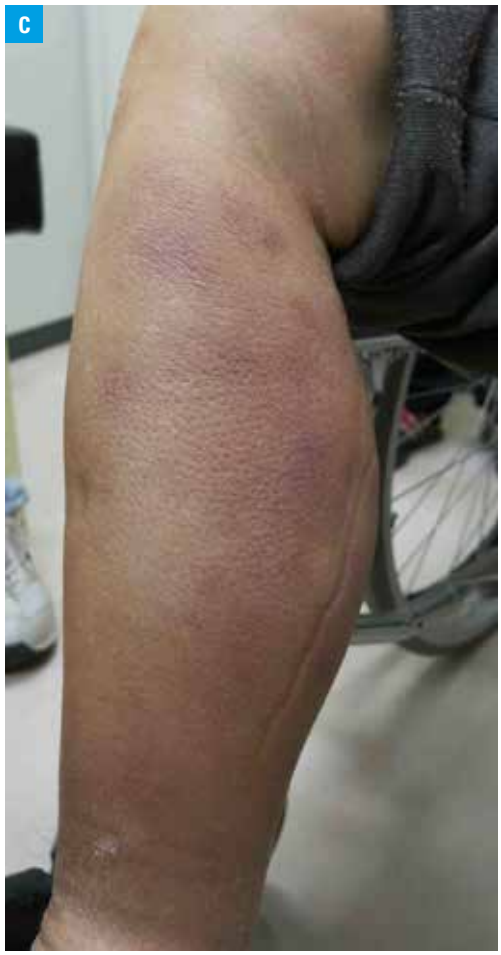

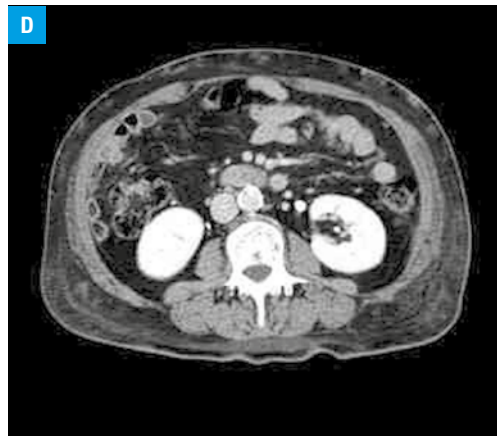

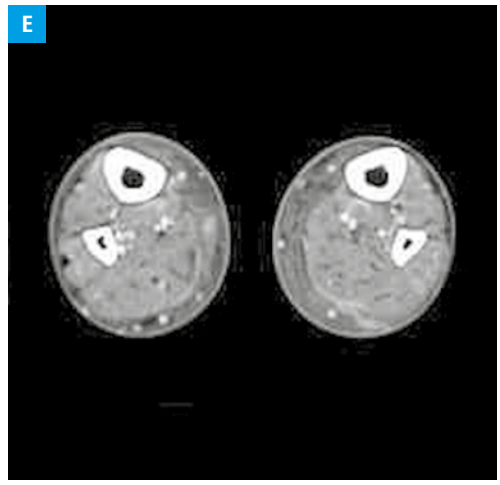

around the blood vessels in the dermoepidermal layer and an intermediate-sized atypical lymphocytic infiltration with rimming of adipocytes in the fat lobules. Immunohistochemistry revealed positive cluster of differentiation (CD) 3, CD4, CD5, CD8, T-cell intracellular antigen-1 (TIA-1), granzyme B, perforin, and high Ki-67 index, as well as negative CD56 staining. Based on ab-T-cell receptor expression analyzed by flow cytometry with CD45 gating, subcutaneous panniculitis-like T-cell lymphoma (SPTCL) was diagnosed. Bone marrow aspiration revealed no evidence of malignancy or hemophagocytic syndrome. Oral cyclosporine (100 mg/d) was started, which resolved the symptoms. The patient showed no relapse over the 18-month follow-up period.

SPTCL is a rare primary cutaneous T/natural killer-cell lymphoma in which cytotoxic T cells infiltrate into subcutaneous adipose tissue without lymphadenopathy. SPTCL commonly develops in young-to-middle-aged people of both sexes, and the clinical presentation includes multiple, painless subcutaneous nodules distributed preferentially on the trunk and lower limbs.[1] Although erythema may appear on parts with aggressive subcutaneous disease progression, there is no infiltration of cytotoxic T cells into the dermis and epidermis. Other symptoms include fever and hepatosplenomegaly. Computed tomography and magnetic resonance imaging reveal multiple diffuse, enhancing, infiltrative subcutaneous lesions or nodules.[2,3] Recently, positron-emission tomography/computed tomography has been reported to be a useful tool.[4] Characteristic pathological, immunohistochemical, and molecular findings on skin biopsy specimens include infiltration of T cells rimming the adipocytes; positive CD3, CD8, TIA-1, and granzyme B and negative CD4, CD30, and CD56 staining; and high Ki-67 index. Immunosuppressive therapy and multiagent chemotherapy are considered. For slow-progressing cases, the first-line immunosuppressive therapy is recommended.[1]

In SPTCL, lesions are likely overlooked because they are painless, diffuse, and occur subcutaneously; therefore, fever and erythema are often the only indicators. While fever and generalized erythema puzzles physicians in general clinical practice, SPTCL should be considered as a differential diagnosis.

REFERENCES

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