Case Report
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Subcutaneous Panniculitis-Like T-Cell Lymphoma in a Patient Receiving Long-Term Panniculitis Treatment: A Case Report

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Abstract

Subcutaneous panniculitis-like t-cell lymphoma (SPTL) is known as a rare type of cutaneous lymphoma characterized with penetration of neoplastic T cells to the subcutaneous tissue. It constituates less than 1% of all non-Hodgkin lymphomas.

We described herein a 49-year-old woman with SPTL who presented with red-purple-colored persistent subcutaneous nodules disseminated on trunk and both extremities. The lesions were present for 15 years. She had been diagnosed as panniculitis and her lesions had not regressed with panniculitis treatment. SPTL was diagnosed due to histopathological examination of the lesions that revealed CD3 positive, CD8 positive, CD4 negative and CD56 negative atypical lymphoid infiltration with lobular and septal panniculitis-like pattern.

SPTL must be always kept in mind as a differential diagnosis, while examining biopsies of patients with panniculitis-like lesions resistant to long-term treatment. It is also important to remember that the biopsy material should be in such depth that involves subcutaneous fat tissue.

Keywords: Panniculitis-like lymphoma, Cutaneous, Lymphoma, T-Cell lymphoma, Histopathology

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Introduction

Subcutaneous panniculitis-like T-cell lymphoma (SPTL) is a rare type of cutaneous lymphoma constituting less than 1% of all non-Hodgkin lymphomas.[1] It is characterized with lobular-pattern penetration of neoplastic α/β T cells to the subcutaneous adipose tissue.[2] SPTL is believed to be a disease of young adults (mean age:36) and is more prevalent among women (male/female ratio: 0.5).[3] SPTL is manifested with red-purple-colored tumoral nodules and plaques usually located deeply in upper and lower

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extremities and trunk, and rarely in the facial area.[2,4,5]

We conducted the present study to discuss an SPTL case with persistent subcutaneous nodules for approximately 15 years, who had been diagnosed as panniculitis, and in whom the lesions had not regressed with panniculitis treatment.

Case Presentation

The 49-year-old female patient was admitted to the Dermatology Outpatient Clinics of our hospital due to red-purple-colored subcutaneous nodules disseminated on her trunk together with her bilateral upper and lower extremities (Figure 1a). Her medical history revealed that these painful lesions, which exacerbated and regressed from time to time for approximately 15 years, had first manifested themselves on her extremities only; however, the lesions progressed to her trunk within the last 4-5 months. The patient had presented for several times to various dermatologists due to these skin lesions. The result of a skin biopsy performed four years ago in another medical facility was reported as panniculitis. Panniculitis treatment was initiated; nevertheless, no regression was observed in the lesions of the patient.

The patient had no symptoms of fever, weight loss, and fatigue. Hepato-splenomegaly, mucosal ulceration, and serous effusion were not identified in the physical examination. Complete blood count, kidney/liver function tests, and viral hepatitis markers were found to be within normal ranges.

Lymphoid infiltration with lobular and septal panniculitis-like pattern was observed in the histopathological examination of the newly performed biopsy (Figure 2). Deep layers of the dermis and subcutaneous fat tissue were found to be infiltrated with medium and small-diameter lymphocytes with narrow cytoplasm. The high-magnification examination revealed lymphocytes with distinct atypical and irregular hyperchromatic

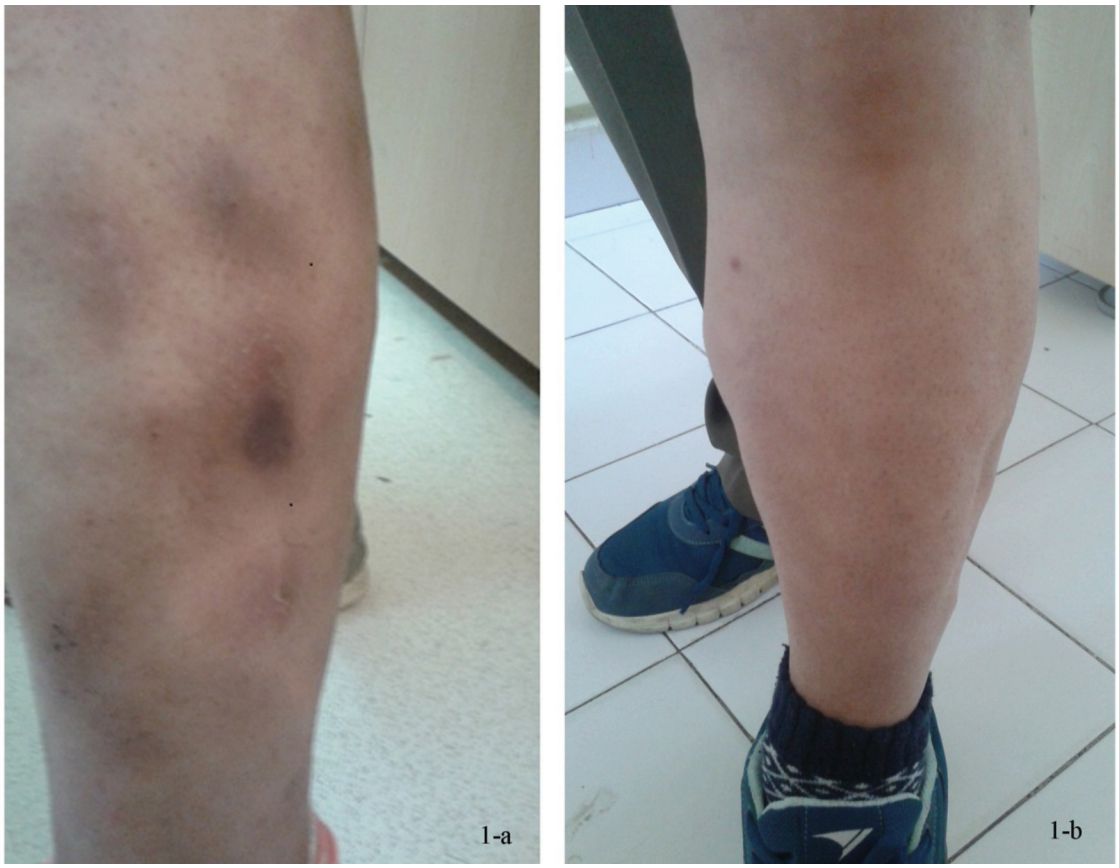


Figure 1. The patient had purplish nodules on her left leg, when she first presented (a). The nodules disappeared after the treatment (b).

nuclei (Figure 3a). Certain atypical lymphocytes surrounded adipocytes (Figure 3b). Necrosis and foamy histiocytes containing cellular fragments and nuclear debris were observed in focal areas (Figure 4). There was no specific presentation in the epidermal and papillary dermal layer. CD3 and CD8 were found to be positive, while CD4, CD20, and CD56 were found to be negative in atypical cells (Figure 5). The patient was diagnosed to have SPTL based on these findings and had consulted the Hematology Department. A patient who had been followed up for three years reported regression of her nodules together with the disappearance of pain and purple-red-colored areas following chemotherapy (Figure 1b).

Discussion

SPTL was initially identified in 1991 by Gonzalez CL. as a new type of T-cell lymphoma, which is similar to panniculitis in terms of clinicopathological features.[6] Even though it had not been classified as a different entity by World Health Organization (WHO) until 2001, it was classified under the group of mature T and NK-cell neoplasia in 2008.[7]

Based on its phenotypic and immune-phenotypic characteristics, SPTL was considered to have two different types as αβ T cell phenotype and γδ T cell phenotype.[8,9] γδ T cell phenotype was excluded from SPTL subgroups and classified as "primary γδ T-cell lymphoma" in 2008 classification of WHO.αβ T cell phenotype was categorized as SPTL.[7] The clinical course of γδ T-cell lymphoma is more aggressive and the risk of hemophagocytic syndrome development increases once compared with αβ phenotype.[10] While the cases with αβ phenotype are usually limited to subcutaneous tissues and have immunophenotypic features of $CD3^+$, $CD4^-$, $CD8^+$, and $CD56^-$, γδ T-cell phenotype has immunophenotypic features of CD3+, CD4-, CD8-, $CD56^+$, and manifests epidermotrophism. Williemze R, Jensen PM et al. considered the α/β and γδ T-cell lymphomas as different entities based on these different characteristics in their study, comprising 83 patients. They calculated the 5-year survival rates of 82% and 11% in

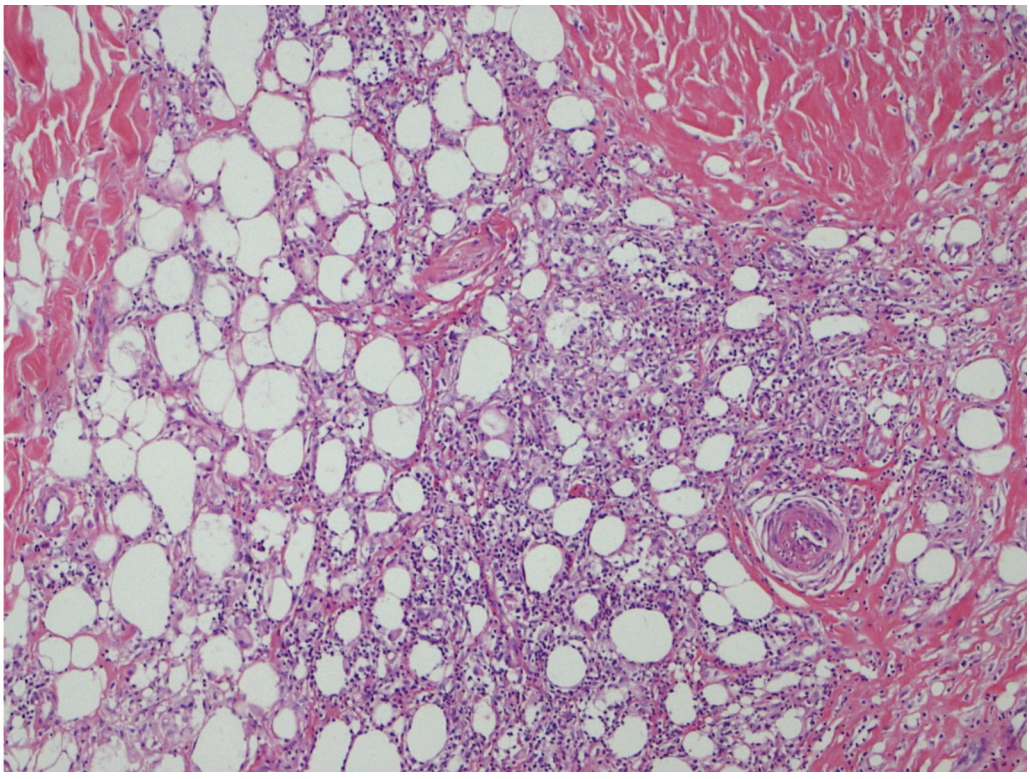
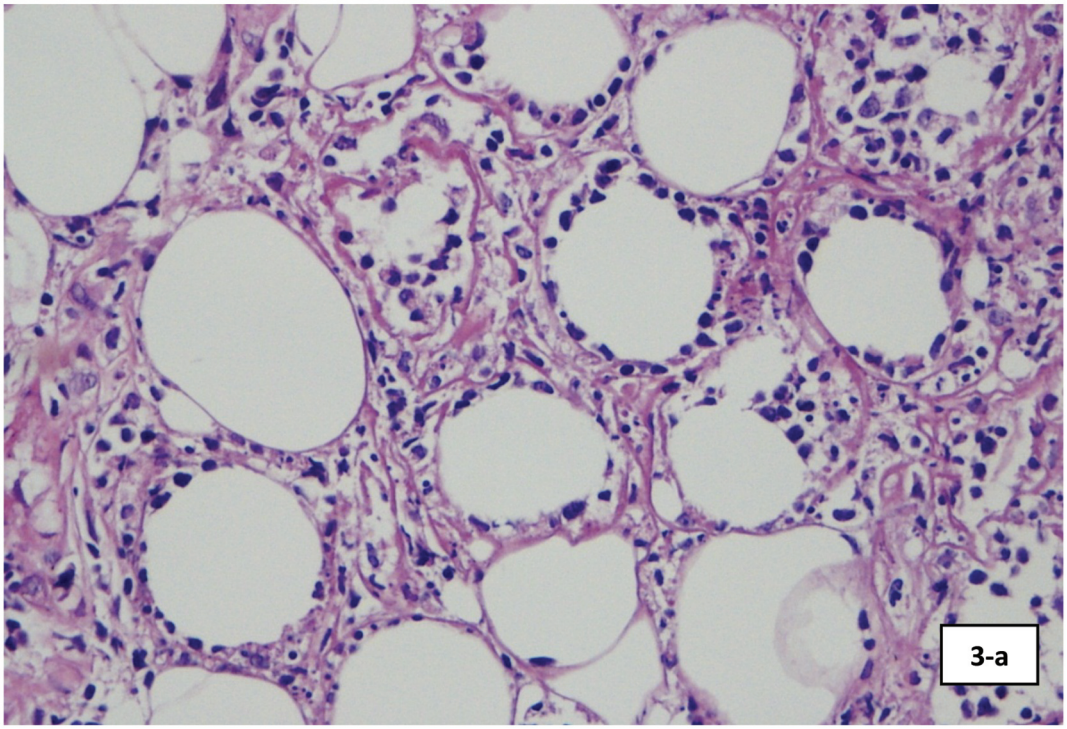


Figure 2. Lymphoid infiltration mimicking panniculitis was seen (H & E, ×100).



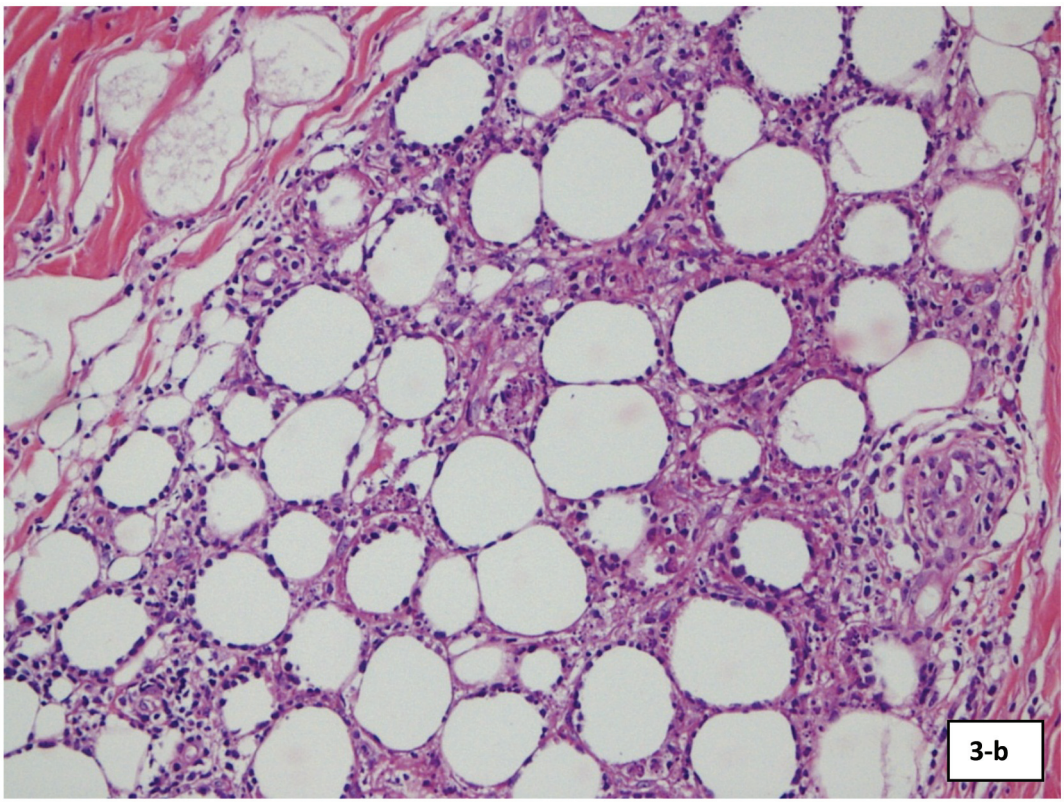


Figure 3. Atypical lymphocytes with irregular hyperchromatic nuclei were seen (H & E, ×400) (a). Some atypical lymphocytes surrounded adipocytes (H & E, ×200) (b).

patients with αβ and γδ T cell lymphomas, respectively.[9]

The histological findings of SPTL are characterized with a panniculitis-like pattern. The infiltration of the subcutaneous area by pleomorphic T-cells and benign macrophages mimics lobular panniculitis. The dermal invasion is minimal and epidermal involvement is rare in general.[5] Lymphocytes have atypical characteristics, such as hyperchromatic angular nuclei and irregular cell borders. Benign histiocytes, plasma cells, and neutrophils may be present. Apoptotic cells, karyorrhectic debris, and focal fat tissue necrosis are usually present.[4,8]

Atypical lymphocytes surrounding adipocytes is an important finding regarding the diagnosis.[4,10] Another important finding could be the large foamy histiocytes containing fat drops, cell fragments, or nuclear debris.[4]

CD3+ and CD4- cytotoxic T-cells are neoplastic cells; these cells are CD8+ and CD56- in general. However, the cells of cutaneous γδ T-cell lymphoma commonly react as CD8- and CD56+.[8,9] Our case was classified as SPTL based on her immunophenotypical characteristics, which could explain that the prognosis was accurate up to an extent even though the patient had not received any specific treatments for 15 years.

In conclusion, we must always keep in mind SPTL as a differential diagnosis, while examining biopsies of patients with panniculitis-like lesions resistant to long-term treatment. It is very important to be careful, while diagnosing panniculitis in such patients. Otherwise, skipping the diagnosis of SPTL could lead to an advanced stage of the disease, as in our case. It is also of great importance to remember that the biopsy material should be in such depth that involves subcutaneous fat tissue.

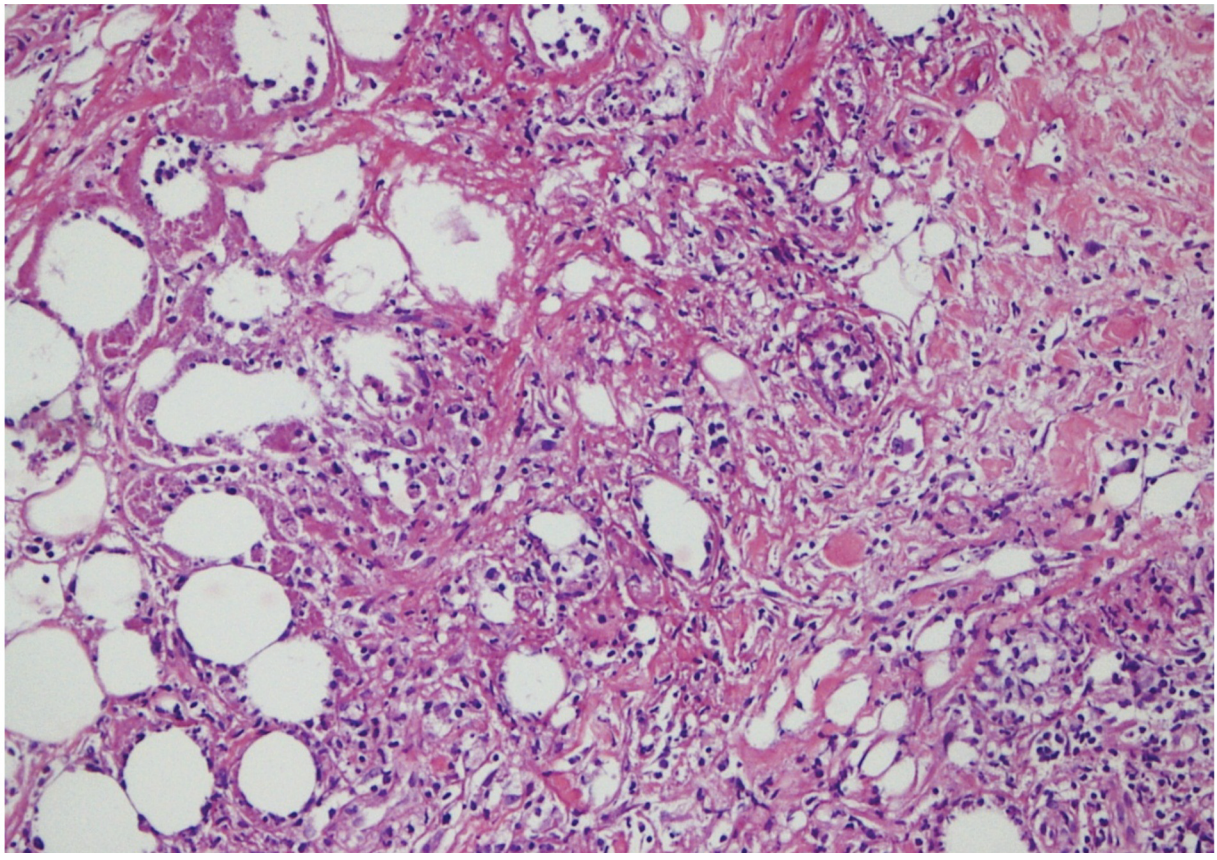


Figure 4. This figure shows necrosis and foamy histiocytes containing cellular fragments (H & E, ×200).

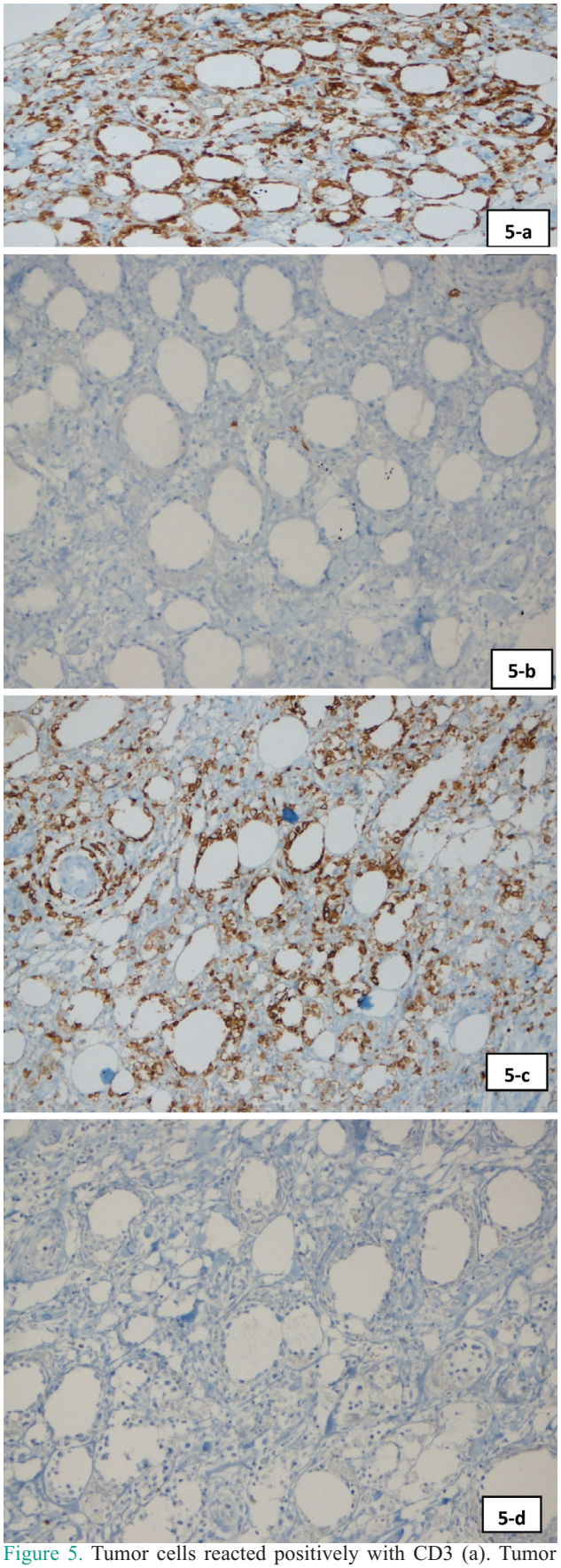


Figure 5. Tumor cells reacted positively with CD3 (a). Tumor cells reacted negatively with CD20 (b). Tumor cells showed CD8 positivity (c). Tumor cells reacted negatively with CD56 (d).

Informed Consent

Patient consent was obtained.

Conflict of Interest

None declared.

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