Relapsed subcutaneous panniculitis-like T cell lymphoma evaluated by FDG PET/CT
A clinical case report
Ping Dong, MDa, Li Wang, MDb, Hongmei Zhu, MDa, Lin Li, MDb,∗

Abstract
Rationale: Subcutaneous panniculitis-like T cell lymphoma (SPTCL) is a rare primary cutaneous T cell lymphomas expressing α/β T cell receptors that preferentially involves subcutis, with a reported proportion of 1% to 2.3% of cutaneous lymphomas.[1–4] As determined by the World Health Organization-European Organization for Research and Treatment of Cancer (WHO-EORTC) classification for primary cutaneous lymphomas, SPTCL was defined as CD8+ cytotoxic T cell lymphoma expressing α/β T cell receptors that are confined to subcutaneous fat, uncommonly associated with hemophagocytic syndrome (HPS).[2,3] While most SPTCL patients will have a relatively indolent clinical course with 5-year overall survival (OS) rate of 82%, some patients presenting with HPS, skin ulceration, or systemic involvement can follow an aggressive course characterized by early relapse.[2]

An accurate diagnosis of relapsed SPTCL is made with a deep skin biopsy that includes subcutaneous tissue (e.g., excisional biopsy) and relies on the constellation of pathologic and immunophenotypic findings.[1,5–7] Several previous studies have demonstrated that FDG PET/CT can be a useful tool for the initial accurate total body staging, restaging following therapy, detecting occult extracutaneous involvement, driving the biopsy towards the most active site, the stratification of prognosis and early therapy assessment.[8–11] To the best of our knowledge, the use of FDG PET/CT in suspicious relapsed SPTCL to clarify the diagnosis has not been previously described. We here report performing FDG PET/CT to explain the diagnosis and monitor post-treatment response of a 15-year-old woman with suspicious relapsed SPTCL.

1. Introduction
Subcutaneous panniculitis-like T cell lymphoma (SPTCL) is a relatively rare subtype of cutaneous non-Hodgkin lymphoma that preferentially involves subcutis, with a reported proportion of 1% to 2.3% of cutaneous lymphomas.[1–4] As determined by the World Health Organization-European Organization for Research and Treatment of Cancer (WHO-EORTC) classification for primary cutaneous lymphomas, SPTCL was defined as CD8+ cytotoxic T cell lymphoma expressing α/β T cell receptors that are confined to subcutaneous fat, uncommonly associated with hemophagocytic syndrome (HPS).[2,3] While most SPTCL patients will have a relatively indolent clinical course with 5-year overall survival (OS) rate of 82%, some patients presenting with HPS, skin ulceration, or systemic involvement can follow an aggressive course characterized by early relapse.[2]

An accurate diagnosis of relapsed SPTCL is made with a deep skin biopsy that includes subcutaneous tissue (e.g., excisional biopsy) and relies on the constellation of pathologic and immunophenotypic findings.[1,5–7] Several previous studies have demonstrated that FDG PET/CT can be a useful tool for the initial accurate total body staging, restaging following therapy, detecting occult extracutaneous involvement, driving the biopsy towards the most active site, the stratification of prognosis and early therapy assessment.[8–11] To the best of our knowledge, the use of FDG PET/CT in suspicious relapsed SPTCL to clarify the diagnosis has not been previously described. We here report performing FDG PET/CT to explain the diagnosis and monitor post-treatment response of a 15-year-old woman with suspicious relapsed SPTCL.

2. Case report
This patient is a 15-year-old woman who received a diagnosis of SPTCL from a thigh skin biopsy 7 years ago. She underwent 12 cycles of chemotherapy and remained asymptomatic without evidence of disease recurrence during her 7-year follow-up until a growing painless subcutaneous mass on perinaeum recurred 2 months ago. Laboratory findings revealed increased aspartate amino transferase and lactate dehydrogenase levels at 73 IU/L (reference range, <40 IU/L) and 259 IU/L (reference range, 110–220 IU/L), respectively. The patient was administered 18F-FDG
and imaged for 2.5 minutes per bed after approximately 60 minutes 18F-FDG injection on a Gemini 16 PET/CT scanner (Philips Healthcare, the Netherlands) for clarifying the diagnosis. FDG PET/CT images demonstrated multiple moderate FDG-avid subcutaneous adipose tissue lesions on the left upper arm (Fig. 1A–D, thin arrows) and perinaeum (Fig. 1A, H–J, arrows), involvement of bilateral inguinal lymph nodes, and a markedly increased FDG-avid subcutaneous mass on the left chest (Fig. 1A, E–G, thick arrows, SUVmax of 5.01), suggestive of relapsed SPTCL. Fortunately, the patient’s skin lesions subsided gradually after 3 cycles of cyclophosphamide, doxorubicin, vincristine, and prednisolone (CHOP) regimen. In addition, a complete remission was observed on interim-FDG PET/CT scan (371.9 MBq) after 3 cycles of CHOP treatment, only with probable inflammatory 18F-FDG activity postchemotherapy on the left chest lesion (Fig. 2E–G, thick arrows, SUVmax of 1.68) without abnormal uptake in other initially involved sites (Fig. 2A–D, H–J, thin arrows and arrows). Extensive cervical brown fat was noted (Fig. 2A, dotted arrows).

This case report was approved by the Ethics Committee of West China Hospital of Sichuan University, Chengdu, China, and the written informed consent was obtained from the patient.

3. Discussion

SPTCL is a rare primary cutaneous T cell lymphoma expressing α/β T cell receptors that preferentially involves subcutis, with an incidence of 1% to 2.3% of cutaneous lymphomas. Compared with other lymphomas involving subcutaneous tissue, such as γδ T cell lymphoma or NK/T cell lymphoma, SPTCL generally shows indolent clinical behavior. However, about 17% of SPTCL patients may develop the HPS, characterized by uncontrolled phagocytosis of blood components, cytopenias, coagulopathy, hepatosplenomegaly, even death. SPTCL patients with HPS had a significantly poorer prognosis than patients without HPS (5-year OS: 46% vs 91%). While most SPTCL patients will have a relatively indolent clinical course, some patients presenting with HPS, skin ulceration, or systemic involvement can follow an aggressive course characterized by early relapse.

An accurate diagnosis of relapsed SPTCL is made with a deep skin biopsy that includes subcutaneous tissue (e.g., excisional biopsy) and relies on the constellation of pathologic and immunophenotypic findings with CD4−, CD8+, CD56−, βF1+ phenotype. Chen et al diagnosed a replapsed SPTCL by performing a skin biopsy again.

The FDG PET/CT imaging features of SPTCL include multiple FDG-avid subcutaneous adipose tissue lesions involving extremities and trunk without a visceral disease. Our case revealed multiple increased FDG-avid subcutaneous mass on the left chest lesion (Fig. 2E–G, thick arrows, SUVmax of 1.68) without abnormal uptake in other initially involved sites (Fig. 2A–D, H–J, thin arrows and arrows). Extensive cervical brown fat was noted (Fig. 2A, dotted arrows).

This case indicated that FDG PET/CT might be considered during clarifying the diagnosis of relapsed SPTCL and detecting more...
occult lesions, avoiding performing skin biopsy again. We recommend performing FDG PET/CT in suspicious relapsed SPTCL to clarify the diagnosis.

**Author contributions**

Data curation: Ping Dong, Li Wang, Hongmei Zhu.
Methodology: Ping Dong.
Resources: Ping Dong, Li Wang, Hongmei Zhu.
Supervision: Lin Li.
Writing – original draft: Ping Dong, Li Wang.
Writing – review & editing: Lin Li.

**References**

[1] Swerdlow SH, Campo E, Pileri SA, et al. The 2016 revision of the World Health Organization classification of lymphoid neoplasms. Blood 2016;127:2375–90.

[2] Willems R, Jansen PM, Cerrolo L, et al. Subcutaneous panniculitis-like T-cell lymphoma: definition, classification, and prognostic factors: an EORTC Cutaneous Lymphoma Group Study of 83 cases. Blood 2008;111:838–45.

[3] Willems R, Jafle ES, Burg G, et al. WHO-EORTC classification for cutaneous lymphomas. Blood 2005;105:3768–85.

[4] Hamada T, Iwatsuki K. Cutaneous lymphoma in Japan: a nation-wide study of 1733 patients. J Dermatol 2014;41:3–10.

[5] Ohtsuka M, Miura T, Yamamoto T. Clinical characteristics, differential diagnosis, and treatment outcome of subcutaneous panniculitis-like T-cell lymphoma: a literature review of published Japanese cases. Eur J Dermatol 2017;27:34–41.

[6] Go RS, Wester SM. Immunophenotypic and molecular features, clinical outcomes, treatments, and prognostic factors associated with subcutaneous panniculitis-like T-cell lymphoma: a systematic analysis of 156 patients reported in the literature. Cancer 2004;101:1404–13.

[7] Parveen Z, Thompson K. Subcutaneous panniculitis-like T-cell lymphoma: redefinition of diagnostic criteria in the recent World Health Organization-European Organization for Research and Treatment of Cancer classification for cutaneous lymphomas. Arch Pathol Lab Med 2009;133:303–8.

[8] Babb A, Zerizer I, Natrek KN, et al. Subcutaneous panniculitis-like T-cell lymphoma with extracutaneous dissemination demonstrated on FDG PET/CT. Am J Hematol 2011;86:375–6.

[9] Rodriguez VR, Joshi A, Peng F, et al. Positron emission tomography in subcutaneous panniculitis-like T-cell lymphoma with extracutaneous dissemination demonstrated on FDG PET/CT. Pediatr Blood Cancer 2009;52:406–8.

[10] Mitsuhashi K, Momose M, Masuda A, et al. Positron emission tomography revealed diffuse involvement of the lower legs and occult extracutaneous lesions in subcutaneous panniculitis-like T-cell lymphoma. Clin Nucl Med 2013;38:209–11.

[11] Wang SY, Wu YW, Hsiao CH, et al. F-18 FDG PET images for subcutaneous panniculitis like T-cell lymphoma. Clin Nucl Med 2011;36:66–9.

[12] Chen CC, Teng CL, Yeh SP. Relapsed and refractory subcutaneous panniculitis-like T-cell lymphoma with excellent response to cyclosporine: a case report and literature review. Ann Hematol 2016;95:837–40.