

Relapsed subcutaneous panniculitis-like T cell lymphoma evaluated by FDG PET/CT

A clinical case report

Ping Dong, MD^a, Li Wang, MD^b, Hongmei Zhu, MD^a, Lin Li, MD^{a,*}

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, and few reports have investigated the diagnosis of suspicious relapsed SPTCL using ¹⁸F-fluoro-2-deoxy-D-glucose (¹⁸F-FDG) positron emission tomography/computed tomography (PET/CT).

Patient concerns: A 15-year-old woman complaining of a growing painless subcutaneous mass on perinaeum recurred 2 months ago, suggestive of suspicious relapsed SPTCL, underwent FDG PET/CT for diagnosis and treatment follow-up.

Diagnosis: Based on the feature of FDG PET/CT images which revealed multiple increased FDG-avid subcutaneous adipose tissue lesions on the left upper arm, the left chest and perinaeum, involvement of bilateral inguinal lymph nodes, and the effective chemotherapy, she was diagnosed with relapsed SPTCL.

Interventions and Outcomes: Fortunately, the patient's skin lesions subsided gradually after 3 cycles of cyclophosphamide, doxorubicin, vincristine, and prednisolone (CHOP) regimen. Besides, complete remission was observed on interim-FDG PET/CT after 3 cycles of CHOP treatment.

Lessons: FDG PET/CT can clarify the diagnosis in suspicious relapsed SPTCL, avoiding performing skin biopsy again.

Abbreviations: ¹⁸F-FDG = ¹⁸F-fluoro-2-deoxy-D-glucose, CHOP = cyclophosphamide, doxorubicin, vincristine and prednisolone, HPS = hemophagocytic syndrome, MIP = maximal intensity projection, OS = overall survival, PET/CT = positron emission tomography/computed tomography, SPTCL = subcutaneous panniculitis-like T cell lymphoma, SUVmax = maximal standardized uptake value, WHO-EORTC = World Health Organization-European Organization for Research and Treatment of Cancer.

Keywords: ¹⁸F-FDG PET/CT, cutaneous T cell lymphoma, subcutaneous panniculitis-like T cell lymphoma

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 aminotransferase 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

Editor: N/A.

The authors have no conflicts of interest to disclose.

^a Department of Nuclear Medicine, ^b Department of Pancreatic Surgery, West China Hospital, Sichuan University, Chengdu, PR China.

* Correspondence: Lin Li, Department of Nuclear Medicine, West China Hospital, Sichuan University, Chengdu, 610041, PR China (e-mail: lilihuaxi@sina.com).

Copyright © 2018 the Author(s). Published by Wolters Kluwer Health, Inc. This is an open access article distributed under the terms of the Creative Commons Attribution-Non Commercial-No Derivatives License 4.0 (CCBY-NC-ND), where it is permissible to download and share the work provided it is properly cited. The work cannot be changed in any way or used commercially without permission from the journal.

Medicine (2018) 97:46(e12980)

Received: 10 June 2018 / Accepted: 4 October 2018

<http://dx.doi.org/10.1097/MD.0000000000012980>

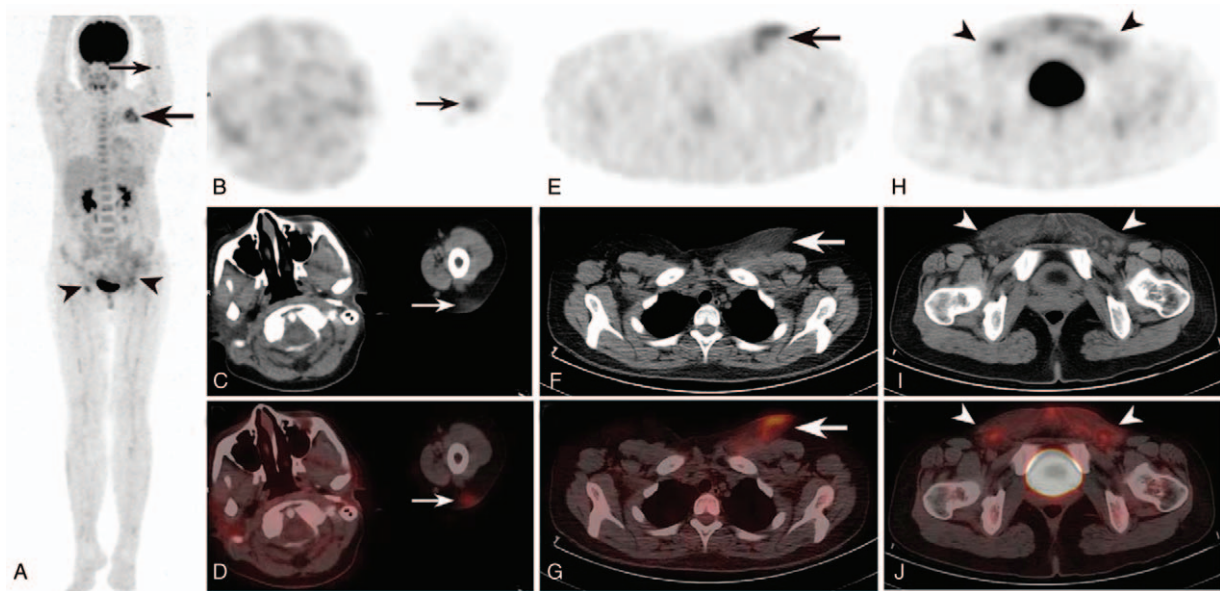


Figure 1. ^{18}F -FDG PET/CT images at baseline of relapsed subcutaneous panniculitis-like T cell lymphoma. FDG PET/CT images [(A) Maximal intensity projection (MIP)]; (B, E, H) PET; (C, F, I) CT; (D, G, J) fusion] demonstrated multiple moderate FDG-avid subcutaneous adipose tissue lesions on the left upper arm [(A–D) thin arrows] and perineum [(A, H–J) arrows], involvement of bilateral inguinal lymph nodes, and a markedly increased FDG-avid subcutaneous mass on the left chest [(A, E–G) thick arrows, SUVmax of 5.01], suggestive of relapsed SPTCL. ^{18}F -FDG = ^{18}F -fluoro-2-deoxy-D-glucose, MIP = maximal intensity projection, PET/CT = positron emission tomography/computed tomography, SPTCL = subcutaneous panniculitis-like T cell lymphoma, SUVmax = maximal standardized uptake value.

(365.2 MBq, 5 MBq/kg body weight) and imaged for 2.5 minutes per bed after approximately 60 minutes ^{18}F -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 perineum (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, maximal standardized uptake value (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 ^{18}F -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.^[1–4] Compared with other lymphomas involving subcutaneous tissue, such as γ/δ T cell lymphoma or NK/T cell lymphoma, SPTCL generally shows indolent clinical behavior.^[3,5] However, about 17% of SPTCL patients may develop the HPS, characterized by uncontrolled phagocytosis of blood components, cytopenias,

coagulopathy, hepatosplenomegaly, even death.^[2,6] SPTCL patients with HPS had a significantly poorer prognosis than patients without HPS (5-year OS: 46% vs 91%).^[2] 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.^[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 with CD4-, CD8+, CD56-, βF1 + phenotype.^[1,2,7] Chen et al^[12] diagnosed a relapsed 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.^[8–11] Our case revealed multiple increased FDG-avid subcutaneous adipose tissue lesions on the left upper arm (Fig. 1A–D, thin arrows), the left chest (Fig. 1A, E–G, thick arrows) and perineum (Fig. 1A, H–J, arrows), with involvement of bilateral inguinal lymph nodes on FDG PET/CT scan. The FDG PET/CT images appear indistinguishable from those due to lobular panniculitis, but are informative in demonstrating disease extension, quantifying disease burden and clarifying the diagnosis of relapsed SPTCL.^[8,9] Several previous studies have demonstrated that FDG PET/CT can be a useful tool for SPTCL the initial accurate total body staging, restaging following therapy, detecting occult extracutaneous involvement, driving the biopsy toward the most active site, the stratification of prognosis and early therapy assessment.^[8–11]

4. Conclusions

This case indicated that FDG PET/CT might be considered during clarifying the diagnosis of relapsed SPTCL and detecting more

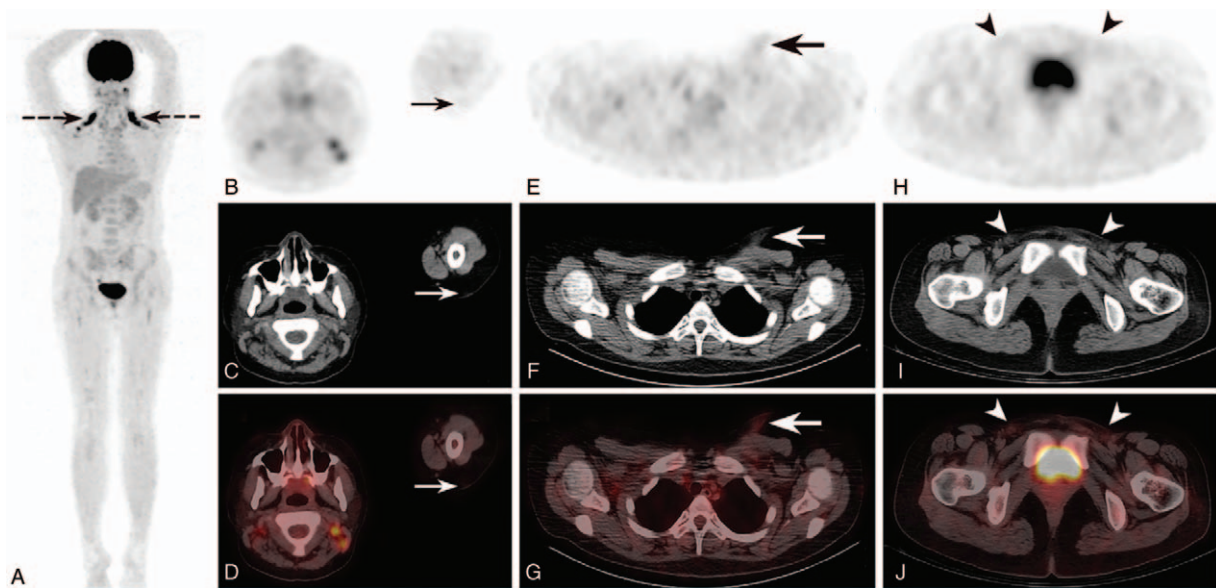


Figure 2. ^{18}F -FDG PET/CT images after CHOP treatment of relapsed subcutaneous panniculitis-like T cell lymphoma. A complete remission was observed on interim-FDG PET/CT scan [(A) MIP; (B, E, H) PET; (C, F, I) CT; (D, G, J) fusion] after 3 cycles of CHOP treatment, only with probable inflammatory ^{18}F -FDG activity postchemotherapy on the left chest lesion [(E–G) thick arrows, SUVmax of 1.68] without abnormal uptake in other initially involved sites [(A–D, H–J) thin arrows and arrows]. Extensive cervical brown fat was noted [(A) dotted arrows]. ^{18}F -FDG = ^{18}F -fluoro-2-deoxy-D-glucose, MIP = maximal intensity projection, PET/CT = positron emission tomography/computed tomography.

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] Willemze R, Jansen PM, Cerroni 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] Willemze R, Jaffe 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, Naresh 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. *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.