Panniculitis: A Rare Presentation of T-Cell Lymphoma in a Tertiary Care Hospital

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Abstract

Subcutaneous Panniculitis-like T-Cell Lymphoma (SPTCL) is a rare and aggressive form of cutaneous T-cell lymphoma that primarily affects the subcutaneous adipose tissue. This study aims to document the clinical presentation, diagnostic challenges and treatment outcomes of SPTCL in a tertiary care hospital setting. Over a period of five years, we reviewed the medical records of patients diagnosed with SPTCL at our institution. The study included a detailed analysis of clinical features, histopathological findings, immunophenotyping and treatment regimens.

The majority of patients presented with multiple, painless subcutaneous nodules on the extremities and trunk. Initial misdiagnoses were common, with many patients initially treated for benign conditions such as panniculitis or dermatitis. Histopathological examination revealed dense infiltration of atypical lymphocytes in a lobular pattern, often rimming adipocytes in a lace-like arrangement. Immunophenotyping confirmed the T-cell origin of the lymphoma, with most cases exhibiting a CD4-CD8+ phenotype.

Treatment approaches varied, with some patients receiving multi-agent chemotherapy regimens, while others were managed with immunotherapy or targeted agents. The study highlights the importance of early and accurate diagnosis, as delayed treatment can lead to significant morbidity and mortality. Despite aggressive therapy, the prognosis of SPTCL remains guarded, with a subset of patients experiencing relapse or progression of the disease.

In conclusion, SPTCL is a rare but serious condition that requires a high index of suspicion for early diagnosis and prompt initiation of appropriate treatment. This study underscores the need for increased awareness among clinicians and the importance of multidisciplinary management to improve outcomes for patients with this challenging lymphoma.

Keywords: Panniculitis • Hemophagocytic syndrome • Immunophenotyping • SPTCL • Lupus erythematosus

Introduction

Subcutaneous panniculitis T-cell lymphoma is a rare slow cytotoxic T-cell lymphoma occurring <1% of all peripheral T-cell lymphoma, with neoplastic α/β T-cell infiltration into the subcutaneous tissue. It presentsas multiple subcutaneous nodules or plaques that occurs in the extremities. They are associated with poor prognosis if compared by hemophagocytic syndrome. Histopathological findings and immunophenotyping are specific of subcutaneous panniculitis T-cell lymphomas and can help in their diagnosis [1].

Case Presentation

A 30-year-old male, recently diagnosed type 2 diabetes mellitus, serology positive (HCV+) came with the complaints of multiple recurrent gradually progressive papulo-nodular swellings all over his abdomen in the last 2 months, initially present over one side of the umbilicus now progressed to the entire abdomen, with mild pain since last 1 month. Associated with recurrent episodes of low-grade fever (39.8°C), night sweats, weight loss. Not associated with pruritus, discomfort. No urinary or bowel complaints (Figures 1 and 2) [2].

Previous history of wound debridement with secondary suturing and IV antibiotics with no remission in symptoms.

On examination: Patient was febrile (39.8°C). Cardiopulmonary system was normal.

Per abdomen: Multiple 1-2 cm painless, diffuse nodular swelling felt over the umbilical, and left lumbar region, which surrounding erythema and induration present. No local rise in temperature or ulcers/sinuses. No generalized lymphadenopathy. No organomegaly [3].

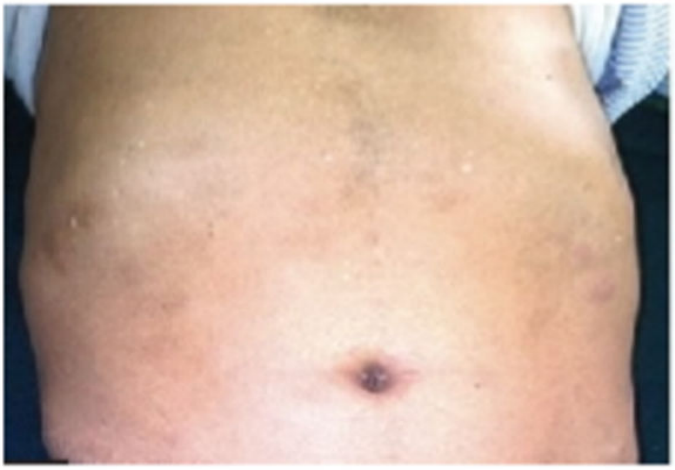

Figure 1. HPE.

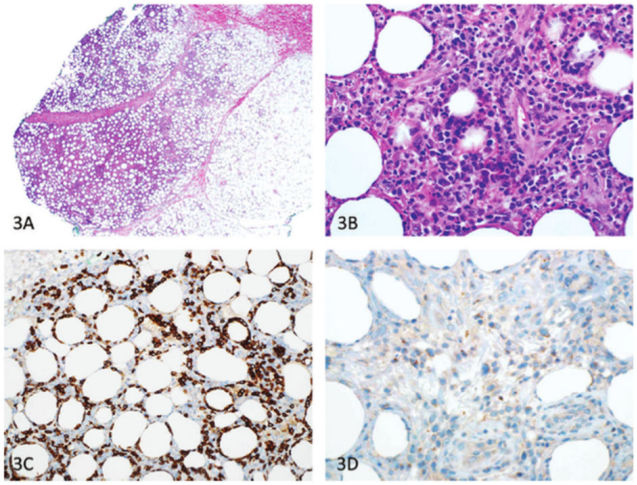

Figure 2. Subcutaneous Panniculitis-like T-Cell Lymphoma (SPTCL). (A) SPTCL showing infiltration of subcutaneous fat lobules by neoplastic cells in a lacelike pattern like that of lobular lymphocytic panniculitis, a finding that may be seen in Lupus Profundus (LP); (B) SPTCL demonstrating rimming of atypical cells around individual adipocytes, another overlapping feature with LP. The majority of atypical cells are CD8 positive; (C) Express cytotoxic markers like TIA-1; (D) (Haematoxylin-eosin, original magnifications 3 20 (A) and 3 200 (B); CD8, original magnification 3 200 (C); TIA-1, original magnification 3 200 (D)). Angioinvasion noted.

Immuno-histomarkers: CD 3(+++), CD2 (+++), CD 5(+++), T-cell Ic antigen-1 (++), Ki67 (+)-25%, CD8(++), perforin +, granzyme +, TIA 1 +CD20(-), CD 30(-), CD79a(-), EBVSRNA (-), CD4(-).

Blood studies: Hb (13.4 gm/dl), TLC-8.03 × 10^3/µL, platelets-236 × 10^3/µL, S. creatinine-0.8 g/dl, Hba1c-11.7%, PET scan (7.3 mCi of 18 FDG).

10 February, 2023 Impression: Enhancing multiple ill-defined hyperattenuating lesions in subcutaneous plane of anterior and lateral abdominal wall, back region, chest wall, gluteal and pelvic regions. No nodal involvement. Show increased FDG uptake/ avidity. SUV Max-9.8.

14 June, 2023 Impression: Normal study.

Treatment: Patient underwent 6 cycles of chemotherapy with CHOP regimen. Follow up PET scan and blood routines were within normal range [4].

Results and Discussion

Subcutaneous panniculitis T-cell lymphoma was initially described by Gonazalez, et al. in 1991 presented 8 cases with subcutaneous panniculitis. It is rare tumor, T-cell origin representing <1% of the non-hodgkin lymphoma, WHO-EORTC, 2001 described SPTCL as a rare tumor that expresses α/β T-Cell Receptors (TCR) gene rearrangement, CD8+, CD4-, CD56-, having a indolent course of recurrent self-healing subcutaneous nodules. Tumor γ/δ TCR are classified as primary cutaneous γ/δ T-cell lymphoma having a rigid clinical deterioration, hemophagocytosis. Incidence of SPTCL is most seen in young children and adult, with high preponderance among females. Median age is 46.5 years, with 20% affecting less than 20 years. SPTCL presents as more than one recurrent painless subcutaneous nodules/indefinite plaques that can affect legs (71%), arms (62%), torso (56%), face (25%) with a diameter of 0.5 cm to 20 cm in size, with presence of necrosis and rarely ulceration. 5-year survival rate is 85%-91% with lesions resolve spontaneously. Upper limb lesions carry the worst prognosis [5].

80% of the SPTCL are symptomatic with associated constitutional symptoms like fever, night sweats, weight loss etc. and bone marrow abnormalities. Most common bone marrow abnormality is Hemophagocytic syndrome which is of high prognostic value. Subcutaneous nodules and plaques have a waxing and waning pattern, deranged LFTs in 50% of the patients.

In a study conducted in Japan, 16 patients with SPTCL presented as B symptoms (81%), hemophagocytic syndrome (45%), autoimmune diseases (13%) with the most common being lupus erythematosus which has been primary disease or can occur simultaneously. The most serious complication of SPTCL is hemophagocytic syndrome which presents as fever, cytopenia, splenomegaly, increased ferritin, deranged LFT, reduced fibrinogen, followed by serosal effusions and pancytopenia. There is similar symptom spectrum between SPTCL and lupus erythematosus and is known as atypical lymphocytic lobular panniculitis. Infiltration of pleomorphic malignant T-cell in subcutaneous adipose tissue with several macrophages is associated with CCR5 regression. Malignant T-cell arrangement around a single fat cell is seen. Clinical diagnosis is difficult due to sensitivity with lupus erythematosus, often simultaneously. The diagnosis of SPTCL is challenging, especially in the early stages when the symptoms mimic other conditions (Figure 3) [6].

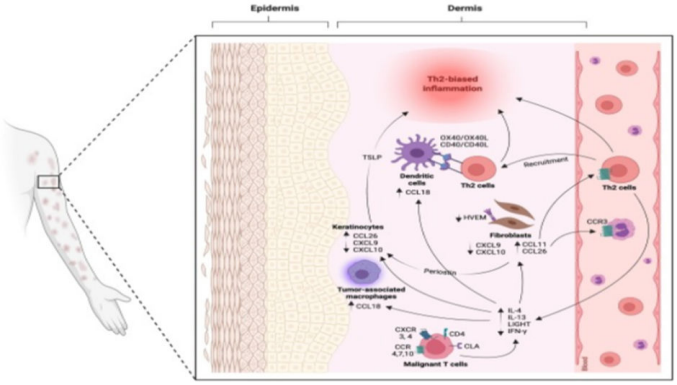

Figure 3. Malignant T-cell arrangement around a single fat cell is seen.

Differential diagnosis

Primary skin α/β T-cell lymphoma: Primarily involves epidermis and dermis, later forming epidermal ulcers. IHC markers are similar CD 56+/TCR-γ. It has the worst prognosis that SPTCL [7].

Extra nodal NK/T-cell lymphoma: Peripheral T-cell lymphoma of NK cell/T-cell line. Association with EBV+. It is associated with mass in the face or skin never restricted to subcutaneous skin. IHC markers CD56+/CD8-/TCR rearrangement (negative) [8].

Benign panniculitis: Subcutaneous fat inflammation. HPE shows no abnormal cells. Clonal TCR rearrangement absent.

Primary cutaneous degenerative large cells lymphoma: Primary T-cell lymphoma is a diffuse infiltration of large lymphoid cells in superficial and deep dermis and subcutaneous tissues. CD3+/IHC-shows expression of cytotoxic molecules T-cell IC antigen-1, granzyme B, perforin, CD30+, CD4+, TCR-γ (Figure 4) [9].

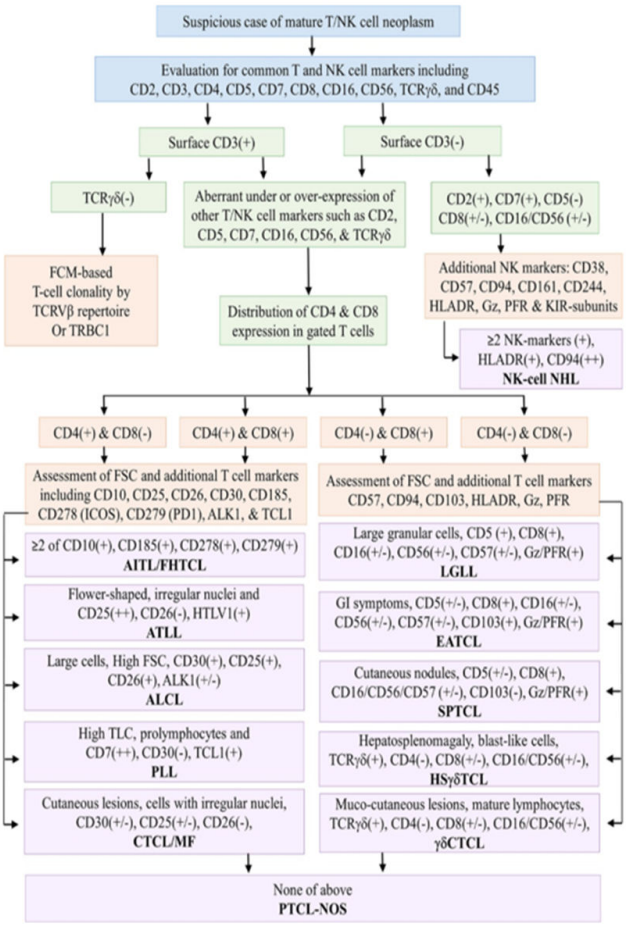

Figure 4. Sucpicious case of mature T/NK cell neoplasma.

Conclusion

Immunosuppressive therapy can result in complete cure with necessity for chemotherapy, sustained low dose hormonal therapy required during the remission period. Systemic corticosteroids/ immunosuppressants such as cyclosporine A, methotrexate, chlorambucil, bexarotene. In case of induced SPTCL attempt with cyclosporine A and dexamethasone is tried. Cyclosporine A with methylprednisolone improves hemophagocytic syndrome in chemotherapy resistant SPTCL with complete remission after 3 months. In both these cases, 1st response has been seen with cyclosporine A. For aggressive disease, doxorubicin based chemotherapy has shown promising results. CHOP regimen cyclophosphamide, doxorubicin, vincristine, prednisolone showing remission rates of 50%. Remission during pregnancy is managed with oral corticosteroids. Radiotherapy is reserved as a palliative measure, thereby inducing late remissions. Stem cell therapy is showing promising results in refractory cases.

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