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CASE SERIES

Subcutaneous Panniculitis-Like T-Cell Lymphoma in Children: Two Case Reports

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Abstract: Subcutaneous panniculitis-like T-cell lymphoma (SPTCL) is a rare primary cutaneous lymphoma derived from cytotoxic αβ T cells, clinically and histopathologically resembling inflammatory diseases of adipose tissue, particularly lupus panniculitis. It accounts for <1% of all non-Hodgkin's lymphomas, with approximately 20% of cases occurring in children. The main aim of this paper was to present two pediatric cases of SPTCL, highlighting the diagnostic challenges involved. The first patient, a 5-year-9-month-old boy, was admitted with a 15 cm infiltrative lesion on the left thigh, previously misdiagnosed and unsuccessfully treated with antibiotics. Imaging revealed an infiltrate resembling lymphedema. A biopsy confirmed SPTCL with a typical immunophenotype. The patient received EURO-LB 02 protocol therapy for peripheral T-cell lymphoma, complicated by pancytopenia, respiratory infection, and polyneuropathy. Post-treatment follow-up showed lesion regression, with residual subcutaneous atrophy (5 cm). The second patient, a 7-year-old girl, presented with a 10 cm inflammatory lesion on the left thigh and systemic symptoms. Imaging and histopathology confirmed the diagnosis. She was treated with the same protocol. Three years later, disease recurrence occurred on the left forearm, managed with alemtuzumab and methotrexate. Both patients remain under outpatient follow-up. Despite its rarity, SPTCL poses a significant diagnostic challenge in children. Accurate differentiation and early diagnosis are crucial for prompt and effective treatment.

Keywords: subcutaneous panniculitis-like T-cell lymphoma, pediatric, children, differential diagnosis, skin diseases

Introduction

Subcutaneous T-cell-like lymphoma (SPTCL) is a rare primary cutaneous lymphoma arising from cytotoxic T αβ cells that clinically and histopathologically mimics adipose tissue inflammation. It accounts for less than 1% of all non-Hodgkin's lymphomas. The median age of onset is 46.5 years, and the disease occurs with similar frequency in men and women. About 20% of cases are diagnosed in patients younger than 20 years.[1] In a retrospective study and review of larger groups of patients with cutaneous lymphomas in children by Alberti-Violetti et al, the incidence of SPTCL was determined to be 6 out of 329 cases of all cutaneous lymphomas.[2] SPTCL manifests as single or multiple subcutaneous nodules, usually reddened, often accompanied by swelling and sometimes ulceration in the center. The lesions are mainly localized on the trunk and extremities, and in 50% of patients coexist with fever, weakness or weight loss.[3] The differential diagnosis includes other T-cell lymphomas and various forms of subcutaneous tissue inflammation, particularly lupus erythematosus (LEP), which accounts for about 20% of all inflammatory cases.[4]

The aim of the following paper was to present the diagnosis and successful treatment of SPTCL based on cases reports.

Case Reports

Case 1

The patient, aged 5 years and 9 months, was admitted to the hospital for an infiltrative lesion involving the skin and subcutaneous tissue of the posterior surface of the left thigh, with a central ulceration (Figure 1A). The diameter of the

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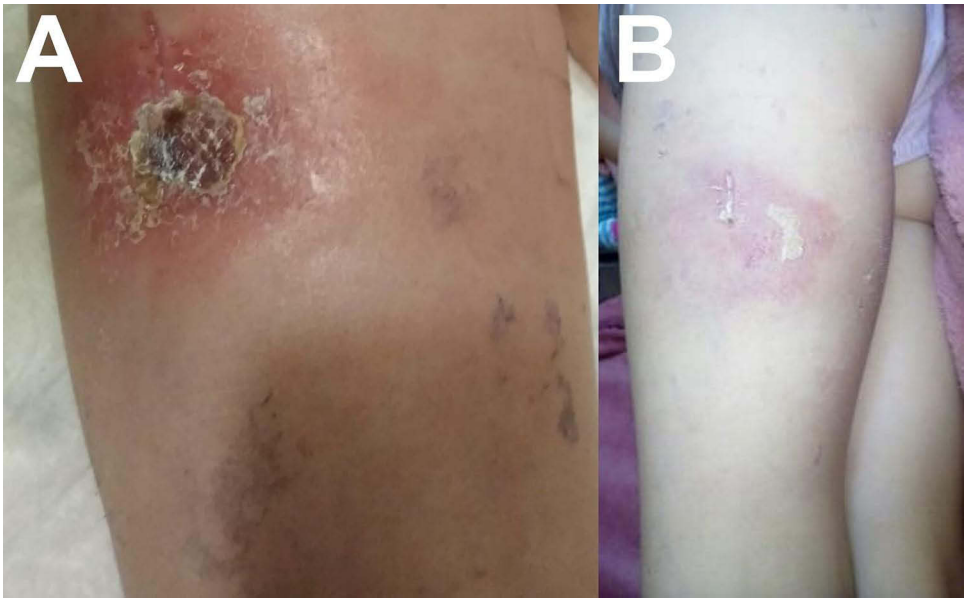

Figure 1 Clinical evolution of the SPTCL lesion on the left thigh: (**A**) before treatment, (**B**) after steroid therapy induction.

lesion was about 15 cm, while the nodular lesion was about 5 cm. Initially, the parents noticed a nodule with a diameter of 5 cm, which rapidly enlarged, taking the form of a hard, longitudinal infiltration (10 cm). The lesion was not painful or itchy. The patient was initially consulted at a surgical and dermatology clinic, where an ultrasound was performed, suggesting inflammation of the subcutaneous tissue. Despite antibiotic therapy, ulceration developed. Because of oncology concerns, a biopsy was performed. Histopathological examination led to a diagnosis of SPTCL (CD3+, CD5-, CD4-, CD8+, CD56+, βF-1(+)). On admission to the Hematology Department, the patient was periodically feverish. Laboratory results showed leukopenia ($3.14x10^3$/μL; N:5–$15x10^3$/μL), elevated lactate dehydrogenase (LDH) level (1118 U/l; N:145–345 U/l]) and slightly elevated transaminases levels (aspartate transaminase [AST] - 47 U/l; N: <42 U/l, alanine transaminase [ALT] - 69 U/l; N:<39 U/l). MRI of the thigh showed an infiltrate with lymphedema morphology, within the described lesions - an indistinct area with reduced signal intensity in T2-weighted images and STIR corresponding most likely to a neoplastic infiltrate, approximate dimension 60×20 x 70 mm (Figure 2). Bone marrow was collected for hematological evaluation. In the myelogram: granulomatous system represented by all transitional forms with a shift to the right to the level of divided granulocyte, in the system draws attention to increased granularity, lymphoreticular system quantitatively below the lower limit of normal). It was decided to implement treatment according to the EURO LB 02 protocol (prednisone, 6-mercaptopurine, methotrexate). The skin lesion after initial steroid therapy is presented in Figure 1B. One month after the start of chemotherapy, MRI of the left thigh was performed again to assess the response to treatment, where a significant regression of the described lesions of the subcutaneous tissue of the left thigh was visualized, the area decreased to 24×7 x 60 mm, the nature of residual edema of the subcutaneous tissue in the area after tumor infiltration (Figure 3). Therapy was complicated by polyneuropathy, bone marrow aplasia, hepatopathy, coagulation disorders and respiratory tract infection. During hospitalization, the patient required broad-spectrum antibiotic therapy, transfusion of blood concentrates and rehabilitation. After 6 months, the patient was admitted to the Hematology Department for anemia (Hb=10.3 g/dl; N:11.5–14.5 g/dl) and thrombocytopenia ($85x10^3$/ul, N:150–400 $x10^3$/μL) after chemotherapy - requiring red blood cell concentrate transfusions. In the follow-up MRI, an oval focal lesion measuring 18×17 x 55 mm, surrounded by a thin sclerotic rim - a picture of bone infarction - was observed in the proximal part of the left femur - in the marrow cavity - a picture of bone infarction, obvious infiltrative lesions in the subcutaneous tissue were not found (Figure 4). After orthopedic consultation, periodic rehabilitation was recommended. The patient continued oncological treatment, which she completed two years after starting therapy. Two-year follow-up after the end of treatment showed the patient's good general condition. Subcutaneous tissue atrophy (5 cm) persisted at the site of the original lesion. There were no significant abnormalities

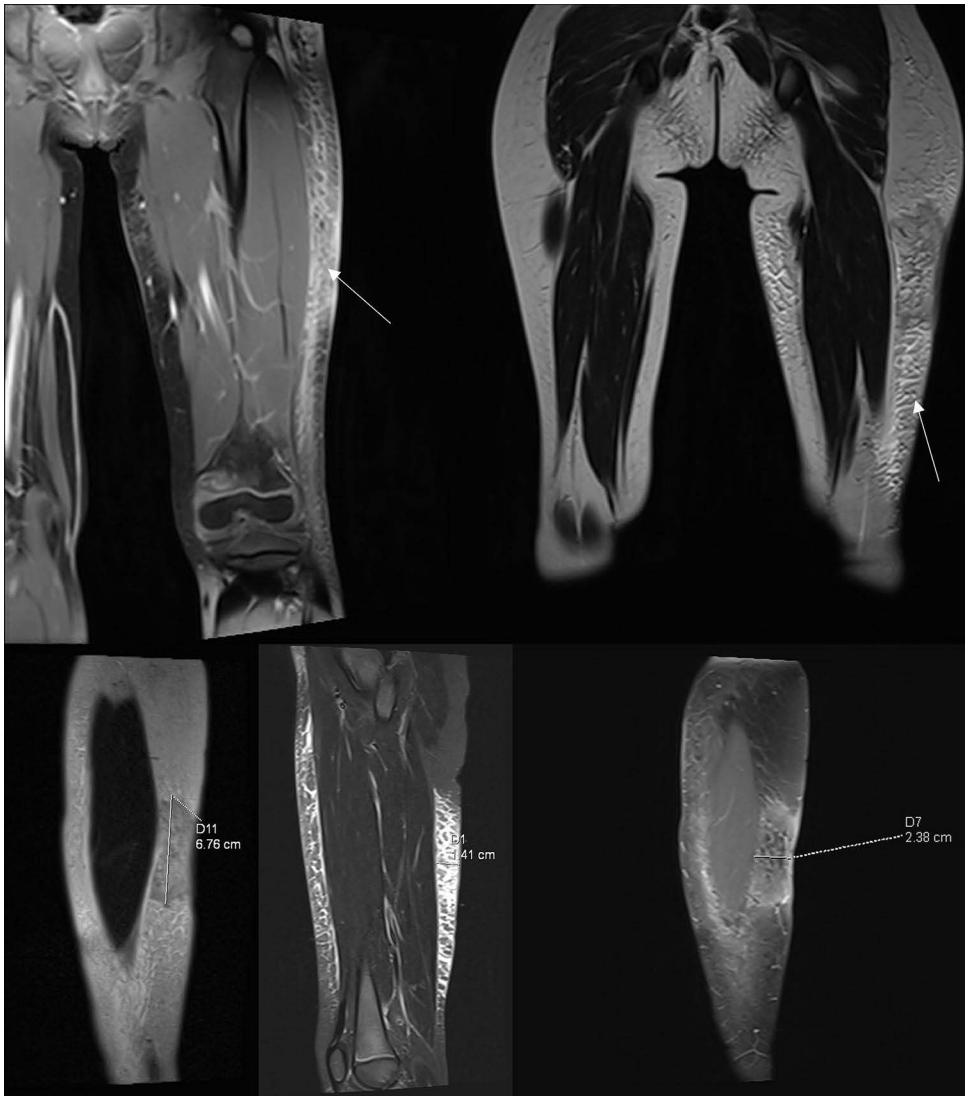

Figure 2 MRI image of the SPTCL lesion of the left thigh before treatment. Infiltrates with lymphoedema morphology, with a diffuse area of reduced signal intensity (60 x 20×70 mm), most likely corresponding to a neoplastic infiltrate. The lesion was measured and marked with a white indicator in the figure.

in the current laboratory results [hemoglobin (14.5 g/dl, N:11.5–14.5 g/dl), leukocytes ($4.72x10^3$/μL; N:5–$15x10^3$/μL) and platelets ($238x10^3$/μL, N:150–450 $x10^3$/μL)].

Case 2

The patient, aged 7 years and 3 months, was admitted to the Clinic because of a massive infiltration of the skin and subcutaneous tissue in the anterior region of the left thigh. Three months prior to the hospitalization, the parents had noticed a small nodule in this area, which gradually enlarged. The changes in the subcutaneous tissue grew, leading to the development of an infiltrate about 10 cm in diameter, with features of inflammation, an erythematous and exudative reaction, a blue-purple border and peripheral oozing. In addition, left inguinal lymph nodes were found to be enlarged. On admission, the patient had a fever of up to 39°C, complained of pain at the site of infiltration, as well as abdominal pain, musculo-articular pain and cough. Laboratory tests showed leukopenia ($1.17x10^3$/μL; N:5–$15x10^3$/μL), anemia (Hb=10.2 g/dl; N:11.5–14.5 g/dl) and elevated ESR (21mm/h; N:3–13mm/h), ferritin (549 ng/mL; N:7–84 ng/mL), lipase (98 U/l; N:<31 U/l) and aminotransferases: AST (53 U/l; N:<42 U/l) and ALT (46 U/l; N:<39 U/l). Broad-spectrum antibiotic therapy was initiated without clinical improvement. Ultrasonography of the inguinal and left thigh showed a nodal conglomerate measuring 24×12 mm with hyperechoic nodal hila. The subcutaneous tissue showed single lymph

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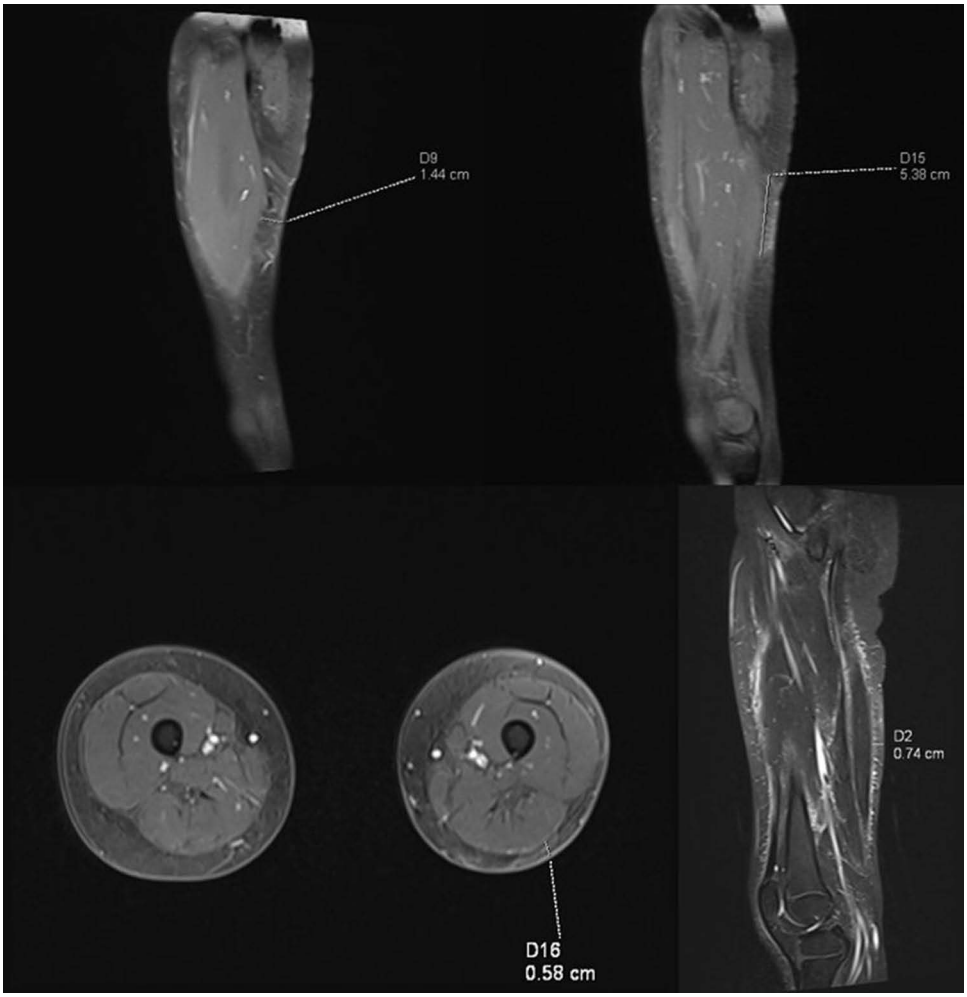

Figure 3 A follow-up MRI showing regression of the primary lesion in one month after the start of therapy. Significant regression of the described lesions of the subcutaneous tissue (24 x 7×60 mm), the nature of residual edema of the subcutaneous tissue. The lesion was measured and marked with a white indicator in the figure.

nodes with reactive features, with a maximum size of 9×4 mm, as well as features of inflammation of the subcutaneous tissue. The skin lesion immediately after skin biopsy is shown in Figure 5. Histopathologic examination of sections taken from the lesion indicated SPTCL, CD3(+), CD4(-), CD8(+), CD52(+) and βF-1(+) (Figure 6). Further gradual progression of the lesion was observed - after one month the infiltrate reached 12×13 cm. Therefore, it was decided to implement chemotherapy according to the EURO-LB 02 protocol, complicated by acute pancreatitis, pancytopenia and respiratory infection. Due to persistent pancytopenia, it was decided to discontinue methotrexate infusions. A follow-up abdominal ultrasound showed a lesion in the spleen, probably of a fungal origin - appropriate antifungal treatment was implemented. Three years after the end of treatment, the patient re-appeared in the Department because of the appearance of a nodule 1.5 cm in diameter on the left forearm, surrounded by an inflammatory infiltrate (10 × 6.5 cm) in the subcutaneous tissue. At the site of the previous lesion on the left thigh, a scar (10 cm) without inflammatory features was visible. The patient was feverish to >38.5°C, laboratory tests showed leukopenia ($3x10^3$/μL, N:4–$10x10^3$/μL). Histopathological examination confirmed recurrence of SPTCL. In the following weeks, new inflammatory infiltrates appeared on both forearms, accompanied by swelling of the left elbow and wrist joint. Ultrasound showed an extensive hyperechoic infiltrate involving the skin and subcutaneous tissue of the left forearm (up to 21 mm thick). Laboratory tests showed persistent leukopenia ($1.83x10^3$/μL, N:4–10 $x10^3$/μL) and elevated CRP levels (4.09 mg/dl, N:<0.5 mg/dl). PET/CT showed metabolically active lesions in the subcutaneous tissue of both upper extremities, a suspicious lesion in the subcutaneous tissue of the left thigh, and enlarged axillary lymph nodes with elevated FDG activity - the disease recurrence was

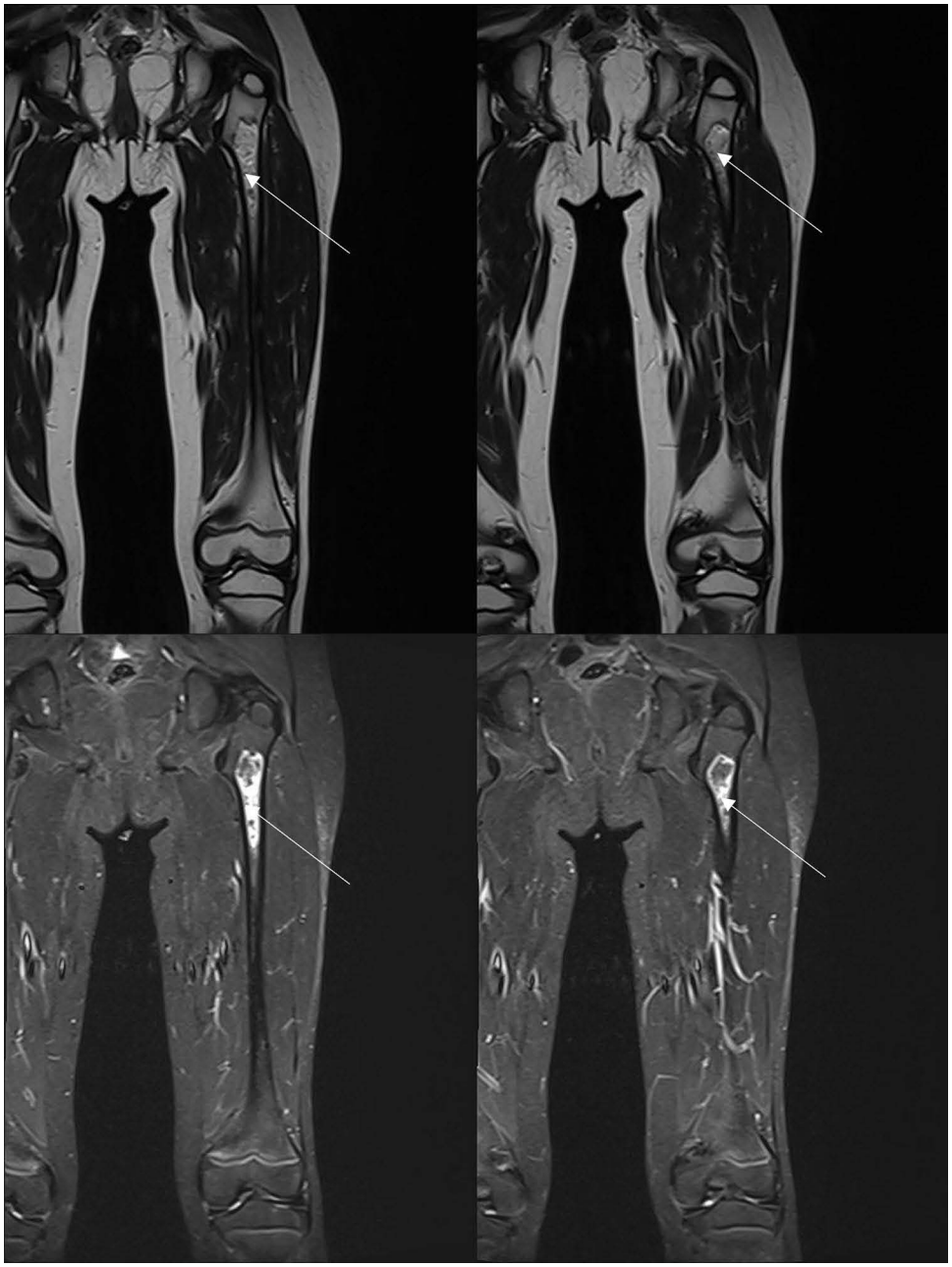

Figure 4 A follow-up MRI in six months after the start of the therapy. In the proximal part of the left femur: oval focal lesion (18 x 17×55 mm), surrounded by a thin sclerotic rim - a picture of bone infarction. The lesion is marked with a white indicator.

confirmed. After consultation with the referring center, treatment with alemtuzumab (0.2 mg/kg, total 9 mg daily over 5 days) and methotrexate was implemented, achieving regression of the lesions. The patient was qualified for allogeneic bone marrow stem cell transplantation (allo-HSCT) from a compatible family donor (brother). Two months after the transplant, the patient was in good general condition, with scarring on her left forearm and left thigh. No recurrence was noted at subsequent follow-up visits.

Discussion

SPTCL, first described in 1991, is an indolent lymphoma manifested by recurrent subcutaneous nodules. It presents an unusual diagnostic challenge due to its nonspecific clinical presentation.[5] Its diagnosis is based mainly on evaluation of biopsy material of the local lesion and proper immunophenotyping. The characteristic phenotype includes: CD3(+), CD4(-), CD8(+), CD56(-)

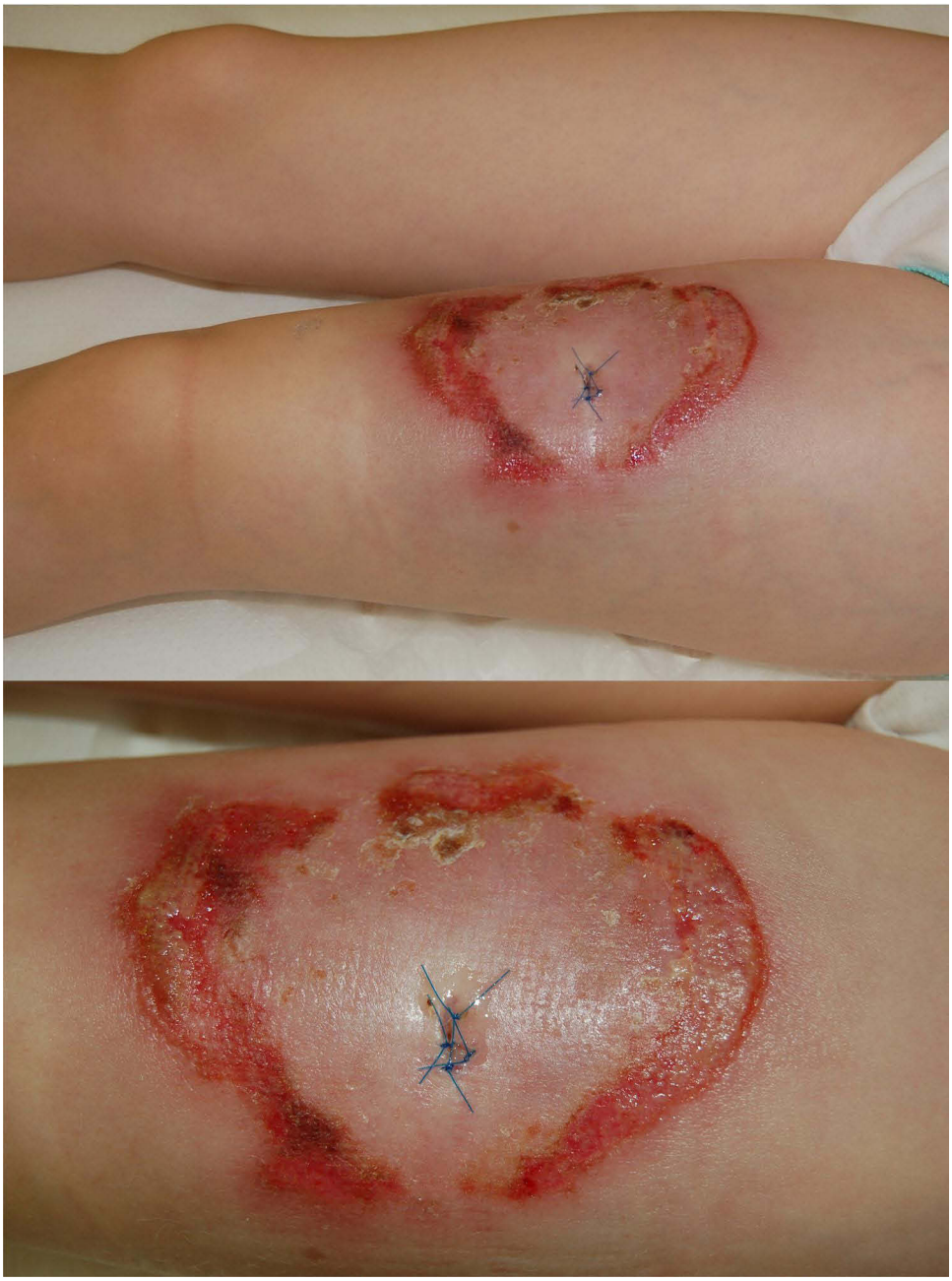

Figure 5 Clinical image of the skin lesion in the patient immediately after the biopsy.

and βF-1(+), as in the patient in question.[5–7] It allows differentiation from the second type - primary cutaneous T-cell γδ lymphoma (PCGD-TCL) with faster progression and frequent coexistence of hemophagocytic syndrome (HLH) (in up to 50% of patients).5,6,7 For PCGD-TCL, the phenotype is more characteristic: CD56(+), perforin(+), granzyme B(+), TIA1(+), CD4 (-), βF-1(-).[6–8] Differentiation from clinically and histologically identical LEP poses a major challenge. It more often occupies the face and upper extremity region, and is also characterized by a low Ki67 proliferation index and mixed inflammatory infiltrates.[9] The above features excluded the diagnosis of LEP in the described patients. Abnormal clonal proliferation of T lymphocytes after stimulation with autoantigen or exogenous antigen has been suggested to be involved in the pathogenesis of the disease.[8] In 25–85% of patients with SPTCL, the *HAVCR2* TIM-3 mutation is found to be responsible for the activation of T lymphocytes and the severe course of the disease.[8] Migration of T lymphocytes into adipocytes is mediated by the affinity of CD5, present on lymphocytes with CCL3, CCL4 and CCL5 present in adipocyte membranes.[10] In the diagnosis of these

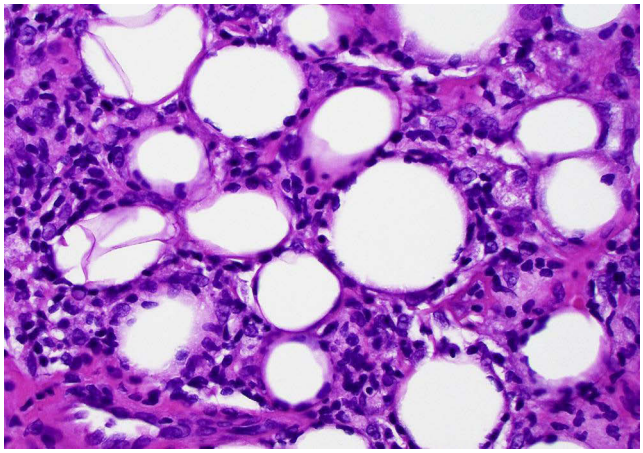

Figure 6 Histopathological image (H+E staining) of a skin biopsy taken from the patient. Lesion showing SPTCL lymphoma with a typical arrangement of lymphocytes surrounding adipocytes (known as rimming).

lesions in children, a whole-body MRI is useful, over a whole-body CT, due to its lower radiation dose.[6,8,11] Due to the rarity of the disease, no universal treatment protocols have been developed.[6–8] Most commonly, as in the present case, polychemotherapy is used, with remission rates of up to 50–67%.[6–11] The preferred regimen is CHOP - cyclophosphamide, doxorubicin, vincristine and prednisolone.[7,8] The above cases demonstrate the efficacy of the EURO-LB 02 protocol, used for lymphoblastic lymphoma and other peripheral T-cell lymphomas.[12] The literature finds other cases of using this regimen with good clinical results, including in infant.[13,14] Most cases of SPTCL, both in adults and pediatric patients, can be effectively treated with immunosuppressive agents.[6–8] Some authors, even recommend that it should be the first-choice treatment.[11] The drug with the highest reported efficacy in this case is cyclosporine A, which is also used to treat relapses.[6–10,15] Other therapeutic options still include bexarotene, chlorambucil and methotrexate.[10] Hematopoietic cell transplantation can also be used in refractory or relapsed cases, as in the second case described.[15] The 5-year overall survival rate in SPTCL is as high as 80%.[6–10] Adverse prognostic factors include the occurrence of HLH (5-year OS of 46%) and involvement of the upper extremity, reported in the second case.[16] The recurrence rate is relatively high at around 30–50%.[8,11,17,18] Youthong et al noted that the recurrence rate was similar in children who received both chemotherapy and immunotherapy as primary treatment (29% vs 37.5%, respectively).[17] Koh et al noted that the presence of the *HAVCR2*^{Y82C} mutation (in 51% of subjects) was associated with more frequent recurrences, with shorter time to progression compared to the general population (11 months vs 30 months, p=0.023).[18] This was associated with significantly more pronounced stimulation of the IL-6-JAK-STAT3 signaling pathway in these patients.[18] There is no consensus on providing intensive therapy for recurrent cases.[15,17] In the case described above, alemtuzumab was used due to the CD52(+) phenotype. Alemtuzumab is a monoclonal antibody directed against cells with this phenotype. It is effective in the treatment of cutaneous T-cell lymphoma (CTCL), especially in relapsed or refractory forms.[19] Awareness of this disease, although rare, is extremely important, as about 30% of pediatric office visits are associated with the presence of skin lesions.[20,21] Their proper differentiation is the basis for therapeutic success.

Conclusion

Subcutaneous T-cell lymphoma is a rare neoplasm with a nonspecific clinical presentation, which can lead to delayed diagnosis. Early histopathological diagnosis is crucial for implementing effective therapy. The cases described here underscore the importance of a multidisciplinary approach in the diagnosis and treatment of hematologic malignancies in children. Alemtuzumab may be effective in treating recurrent disease.

Data Sharing Statement

All data are included in the paper.

Ethical Statement and Informed Consent

Written informed consent for publication of case details and accompanying images was provided by the patients' parents. In accordance with the rules at the Medical University of Lublin, the consent of the Head of the Department of Paediatric Haematology, Oncology, and Transplantology, Medical University of Lublin for the publication of this paper has been obtained.

Author Contributions

All authors made a significant contribution to the work reported, whether that is in the conception, study design, execution, acquisition of data, analysis and interpretation, or in all these areas; took part in drafting, revising or critically reviewing the article; gave final approval of the version to be published; have agreed on the journal to which the article has been submitted; and agree to be accountable for all aspects of the work.

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Disclosure

The authors report no conflicts of interest in this work.

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