Abstract The case of a 40-year-old black man, who developed a very unusual tumour-like lymphoid hyperplasia involving primarily the subcutaneous tissue, is reported. The lesion, which arose at a site of tribal scarifications, displayed a deceptive morphology that closely resembled subcutaneous panniculitis-like T-cell lymphoma (SPTCL). An accurate diagnosis could only be made following detailed immunohistochemical and molecular studies. Although SPTCL has been thought to represent a very specific clinicopathologic entity, the present case illustrates that its histological appearance can, however, be closely mimicked by reactive and benign conditions.

Keywords Subcutaneous panniculitic T-cell lymphoma · Pseudolymphoma · Subcutis · Scarification

Introduction Various inflammatory diseases of the skin may resemble cutaneous lymphomas clinically and/or histologically [10]. Depending on the predominant cell type that makes up the infiltrate, these pseudoneoplastic conditions have been divided into T- and B-cell pseudolymphomas, respectively [2, 10]. They mostly involve the dermis and, at times, may extend to the subcutaneous fat. However, lymphoid hyperplasia, arising exclusively in the subcutaneous tissue, remains distinctly rare [4, 6]. Subcutaneous panniculitis-like T-cell lymphoma (SPTCL) is a distinct neoplasm that usually follows an aggressive clinical course. This rare tumour preferentially involves the subcutaneous tissue of the extremities and/or of the trunk, and a significant number of cases are complicated by a haemophagocytic syndrome [3, 7, 11, 14, 16]. Histologically, this neoplastic disorder is made up primarily of small- or medium-sized lymphocytes that involve the subcutaneous fat in a way very similar to that of a lobular panniculitis [7, 11, 14, 16]. When investigated using immunohistochemistry, most SPTCL cases show a CD3+, CD4–, CD8+, CD56– T-cell phenotype and consistently express markers of cytotoxic T lymphocytes, such as cytolytic granule-associated protein TIA-1, perforin, or granzyme B. In this context, rimming of fat spaces by CD8-positive lymphocytes is usually considered as a characteristic feature of SPTCL [2]. With regard to the T-cell receptor (TCR) phenotype, the αβ TCR subtype is more frequently found than the γδ TCR one [11]. On molecular grounds, TCR gene rearrangement analysis usually demonstrates clonal rearrangements of the genes coding for the β, the γ, or the δ subunits of the TCR [7, 11, 14].

Herein, we report a very unusual form of lymphoid hyperplasia, primarily involving the subcutis and, by morphology, closely mimicking SPTCL. In addition, this lesion arose at a site of tribal scarifications, therefore suggesting a possible relationship between the two conditions.

Case report A 40-year-old black man, originating from Benin, was referred to the hospital because of a tumour mass of the right cheek. The tumour mass, which spontaneously appeared 6 weeks before, was characteristically localised under ethnic scars. These tribal scarifications were made in the youth, and the patient had no significant medical history apart from a positive serology for hepatitis A virus and a primary tuberculosis infection, 10 years before. Neither recent drug intake nor arthropod bite were recorded. Laboratory findings didn’t disclose any significant alterations with the exception of a slight increase in lipase serum levels (43 mU/ml). Anti-nuclear antibody (ANA), rheumatoid factor, and antineutrophil cy-
toc acid (EDTA), 0.45% Tween20] for 48 h at 37°C. For the IgH
in buffer [50 mM Tris (pH 8.5), 1 mM ethylene diamine tetraace-
tive ISH detection system (BioGenex, San Ramon, Calif.). The
EBV-encoded RNA (EBER) was performed, using a supersensi-
Glostrup, Denmark; CD5 and CD57 Novocastra Laboratories Ltd,
CD3, CD8, CD20, CD21, CD35, CD45RO and CD79a, Dako A/S,
and a new biopsy was performed.

Materials and methods
Formalin-fixed tissue samples from the initial lesion and its recurrence were routinely processed and embedded in paraffin, accord-
ing to standard histological techniques. Sections were stained with haematoxylin and eosin. Immunohistochemical studies were per-
formed on paraffin sections using a Ventana Nexes Staining System (Ventana Medical Systems, Tucson, Ariz.). Antibodies di-
rected against the following antigens were used: CD43, BioGenex, San Ramon, Calif.; κ and λ immunoglobulin (Ig) light chains, CD3, CD8, CD20, CD21, CD35, CD45RO and CD79a, Dako A/S, Glostrup, Denmark; CD5 and CD35 Novocastra Laboratories Ltd, Newcastle-upon-Tyne, UK; granzyme B: Monosan, AM-Uden, NL; CD30: NeoMarkers, Union City, Calif.; TIA-1, Immunotech SA, Marseille, France.

In order to investigate the possible role of EBV infection in the pathogenesis of this disorder, an in situ hybridisation (ISH) for EBV-encoded RNA (EBER) was performed, using a supersensi-
tive ISH detection system (BioGenex, San Ramon, Calif.). The search for clonal Ig heavy chain (IgH) gene rearrangement was performed using the polymerase chain reaction (PCR) analysis [12]. For this purpose, formalin-fixed paraffin-embedded tissue from the lesions was used. Briefly, five 10-µm thick sections were dewaxed and subsequently digested with proteinase K (200 µg/ml) in buffer [50 mM Tris (pH 8.5), 1 mM ethylene diamine tetraace-
tic acid (EDTA), 0.45% Tween20] for 48 h at 37°C. For the IgH
PCR analysis, consensus primers complementary to the IgH framework 3 region (5'-CTGTCGACCCGGGTATTACCG-
γ1.4-cm tumour process involving the outer
part of the cheek’s wall (Fig. 1). Complementary clinical investi-
gations and systemic image studies failed to reveal systemic in-
volve. Therefore, a surgical but incomplete excision was per-
formed for diagnostic purposes. Since the lesion was considered
as probably benign, no further therapy was given. The facial nod-
ule locally recurred and increased in size 10 months after the ini-
tial diagnosis. There was still no evidence of disseminated disease, and a new biopsy was performed.

Pathological findings
Pathological study of the initial lesion and that of the re-
currence revealed similar findings. Both tumours were composed of lymphoid cells of heterogeneous morpholo-
gy involving the subcutaneous tissue (Fig. 2 and Fig. 3). The pattern of adipose tissue infiltration typically resembled that of a lobular panniculitis with some myxoid de-
generation of the adipocytes. There was also some vas-
cular hyperplasia. At higher magnification, the cellular infiltrate was composed of a few large or medium-sized cells, scattered among numerous small lymphocytes, some plasma cells, and histiocytes. The latter occasionally-
ly exhibited features of erythrophagocytosis (Fig. 4) but didn’t cluster together to form granulomas. In addition, some areas featuring karyorrhectic bodies were also no-
ticed (Fig. 5).

The overall immunophenotypic results visualised relatively distinct B-cell areas accompanied by a T-cell population. The B-cell component, recognised by the expression of CD20 and CD79a, comprised a majority of small lymphocytes and a minority of larger cells. CD21 and CD35 immunostainings occasionally showed the presence of follicular dendritic cells in some of