Palpable Archiform Migratory Erythema Preceded by B-cell Pseudolymphoma in a Japanese Man

Hiroaki Yagi, Taisuke Ito, Natsuho Ito, Takahiro Horibe, Yasushi Yoshinari, Masahiro Takigawa and Hideo Hashizume
Department of Dermatology, Hamamatsu University School of Medicine, 1-20-1 Handayama, Hamamatsu 431-3192, Japan. E-mail: hiroyagi@hama-med.ac.jp
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Sir,

Palpable archiform migratory erythema (PAME) is a rare member of the family of T-cell pseudolymphomas occurring preferentially in adult males (1–5). The clinical picture of PAME is distinctive from other pseudolymphomas because infiltrated annular erythema develops into elevated migrating lesions in the trunk as the predilection site. The aetiology of this condition is unknown and the disease runs a chronic course. We report here a case of PAME in which polyclonal populations of T cells proliferated surrounding a preceding B-cell pseudolymphoma.

CASE REPORT

A 66-year-old man had developed a red, small lump 10 years previously and after 7 years arc-shaped eruptions began to appear surrounding this initial lesion. The initial examination revealed an indurated smooth surfaced nodule 2.5 cm in diameter surrounded by archiform and annular, firm, palpable erythemas on the back (Fig. 1a). There were no subjective symptoms or superficial lymphadenopathy. The central nodule gradually resolved in 3 months after the first visit to our clinic without any treatment. The archiform and annular erythemas waxed and waned, as a part of the lesions disappeared in 2–3 months while other parts tended to clear, without any systemic or topical treatment (Fig. 1b). While oral penicillin, topical steroids of strong potency, and intra-lesional and intravenous injections of interferon-γ (6) were ineffective, oral administration of prednisolone at a starting dose of 30 mg/day followed by dose-tapering completely cleared the lesions in 8 months.

Laboratory findings including red and white blood cell counts, haemoglobin levels, serum electrolytes, liver enzyme levels, serum protein and immunoglobulin levels and C-reactive protein were normal on several occasions during our 3-year observation. Anti-nuclear antibodies, and antibodies to *Treponema pallidum*, human T-lymphotropic virus-I, human immunodeficiency virus, hepatitis C virus, and *Borrelia burgdorferi* were always negative. Chest X-ray showed no abnormality, and there was no lymphadenopathy or hepatosplenomegaly by computerized tomography scanning. Bone marrow aspiration revealed normal cellular composition.

In the central nodule, atypical lymphoid cells with large, irregularly shaped, pale nuclei and small lymphocytes lay either intermingled with one another or in a follicular arrangement in the entire upper dermis (Fig. 2a). Lymphoid follicular structures consisted of $CD20^+$ large atypical B and $CD3^+$ small T cells at a ratio of 9:1 (Fig. 2b). At resolution, mononuclear cell infiltrates localized to perivascular areas with less conspicuous follicular structures that consisted of almost equal numbers of $CD20^+$ and $CD3^+$ cells. Specimens obtained from arciform erythema on 3 occasions at different sites consistently revealed a dense lymphocytic infiltrate around the blood vessels and hair follicles in the upper and mid dermis, with a $CD3^+$ to $CD20^+$ cell ratio of 7 to 3 (Fig. 3). No follicular structure was apparent. In addition, helper T cells outnumbered cytotoxic T cells since $CD4^+$ and $CD8^+$ cells always comprised 60–80% and 20–40% of T-cells, respectively. $CD4^+$ cells in the perivascular infiltrate expressed either CCR4 (Th2 cells) or CXCR3 (Th1 cells) (7). Southern blot analyses on several occasions during our 3-year observation showed polyclonal proliferation of T and B cells in the central nodule and arciform erythema.

Alcian blue staining showed no deposition of mucin in the dermis and subcutis, and lupus band tests were negative in both types of lesions. Thus, clinical pictures, histology, and immunohistochemistry indicated that the nodule and archiform erythemas represent lymphocytoma cutis and PAME, respectively.

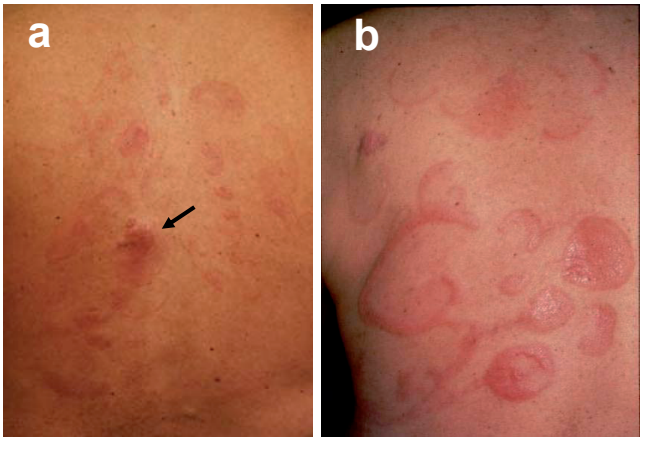

Fig. 1. Different time-points of the clinical manifestation on the back. (a) Firm nodule (*arrow*) surrounded by archiform, raised erythemas at the first visit. (b) Migrating archiform erythemas that waxed and waned during 6 months.

DISCUSSION

T-cell pseudolymphomas including PAME are characterized by the proliferation of benign hyperplastic T cells, mostly of the $CD4^+$ phenotype (2–4, 8, 9), with the exception being CD8-dominant PAME in an HIV-infected individual (5). Although the aetiology is unknown in most cases, T cells persistently accumulate and proliferate in response to a variety of stimuli. The high proportion of $CD8^+$ T cells among lymphocytic infiltrate in $CD4^+$ T-cell pseudolymphoma may reflect an effective host immune response against proliferating $CD4^+$ cells, which prevents further progression (10). A unique clinical feature of our patient was that PAME developed surrounding preceding B-cell pseudolymphoma. It is possible that the prolonged interaction of infiltrating T cells with these pseudolymphomatous

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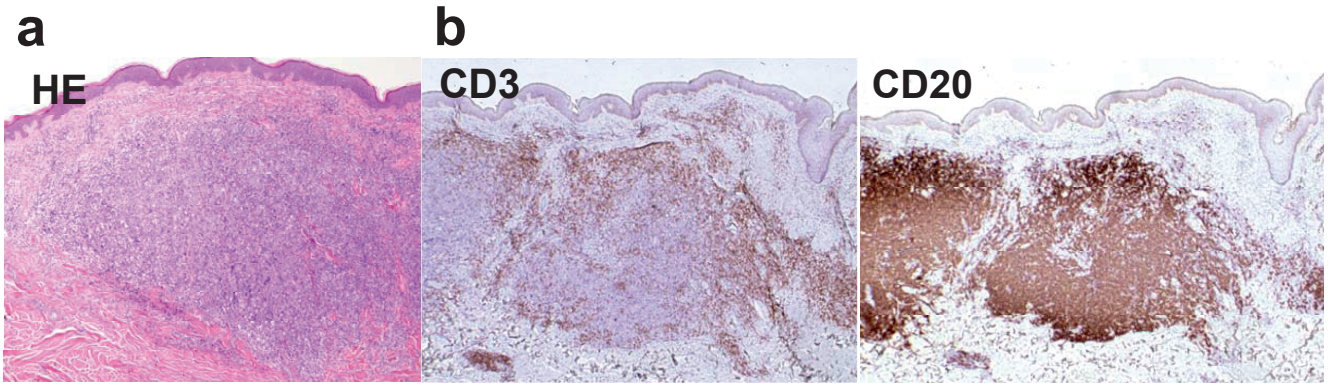

Fig. 2. Routine histology and immunohistochemistry of specimen from a nodule. (a) H&E staining showing lymphoid follicles in the upper dermis (×100). (b) CD3 and CD20 expression (×100).

B cells resulted in hyperplasia of Th1 (CXCR3) and Th2 (CCR4) cells as a clinical manifestation of PAME. The persistence of archiform eruptions even after the disappearance of lymphocytoma cutis suggested an ineffective but protracted protective immunity against pseudolymphomatous B cells.

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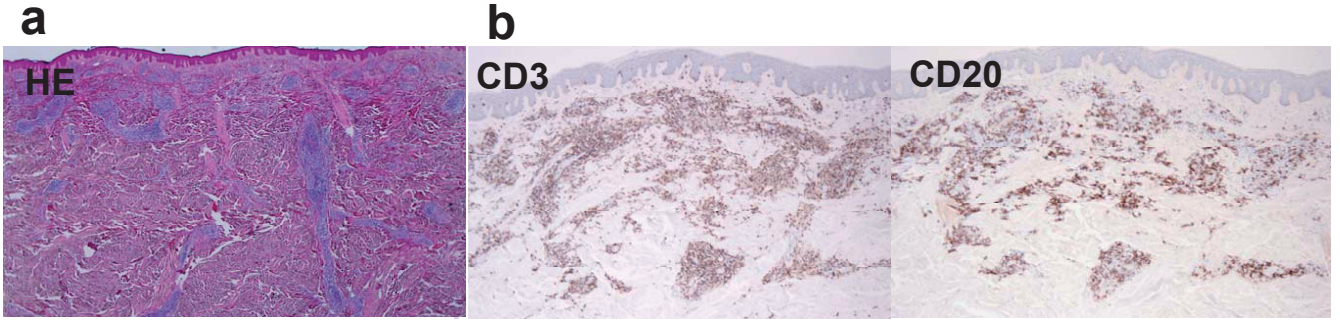

Fig. 3. Routine histology and immunohistochemistry of specimen from palpable archiform erythema. (a) H&E staining (×100). (b) CD3 predominance over CD20 (×100).