REFERENCES

1. Kamra HT, Gadgil PA, Ovhal AG, Narkhede RR. Steatocystoma multiplex-a rare genetic disorder: a case report and review of the literature. J Clin Diagn Res 2013;7:166-168.
2. Jeong SY, Kim JH, Seo SH, Son SW, Kim IH. Giant steatocystoma multiplex limited to the scalp. Clin Exp Dermatol 2009;34:e318-e319.
3. Liu Q, Wu W, Lu J, Wang P, Qiao F. Steatocystoma multiplex is associated with the R94C mutation in the KRTl7 gene. Mol Med Rep 2015;12:5072-5076.
4. Covello SP, Smith FJ, Sillevis Smitt JH, Paller AS, Munro CS, Jonkman MF, et al. Keratin 17 mutations cause either steatocystoma multiplex or pachyonychia congenita type 2. Br J Dermatol 1998;139:475-480.
5. Irvine AD, McLean WH. Human keratin diseases: the increasing spectrum of disease and subtlety of the phenotype-genotype correlation. Br J Dermatol 1999;140:815-828.

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A Case of Mycosis Fungoides Occurring after Primary Gamma-Delta T-Cell Lymphoma

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Dear Editor:

Mycosis fungoides (MF) represents the most common type of cutaneous T-cell lymphoma, characterized by epidermotropic proliferation of small-to medium-sized T-cells[1]. On the other hand, primary cutaneous gamma-delta T-cell lymphoma (PCGD-TCL) is composed of a clonal proliferation of mature, activated gamma-delta T-cells with a cytotoxic phenotype[1]. Here, we report a case of MF following PCGD-TCL. To our knowledge, this is the first reported case.

A 52-year-old woman presented with a 2-year history of erythematous lesion with crust and erosion on the right pubic area. She had no particular medical history. A biopsy specimen revealed infiltration of medium to large sized atypical lymphoid cells in the dermis and subcutis (Fig. 1). We received the patient's consent form about publishing all photographic materials. The majority of atypical cells were CD3+, CD4−, CD8−, and CD56+. Also immunostaining revealed the absence of T-cell receptor (TCR)-βF1. Laboratory examination revealed leukocytosis (9,900/μl; normal: 3,150∼8,630/μl). Positron emission tomography showed no internal organ involvement. The patient was diagnosed with PCGD-TCL. The patient received multidrug chemotherapy; 6 cycles of bortezomib, cyclophosphamide, adriamycin, vincristine, and prednisolone. After 1 year PCGD-TCL recurred and the patient received radiotherapy after 6 cycles of chemotherapy. This resulted in complete response for PCGD-TCL. Three years later, the patient revisited with erythematous scaly patches on her extremities and trunk. A biopsy specimen showed epidermal and dermal infiltration of atypical lymphocyte (Fig. 2). Phenotypes of these cells were CD3+, CD4+,

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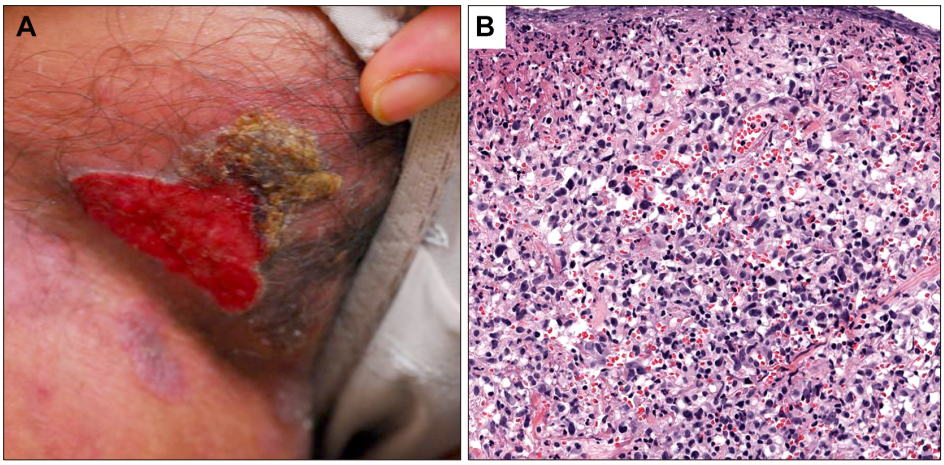

Fig. 1. (A) Erythematous patch with ulcer and crust on the right pubic area. (B) Infiltration of medium to large sized atypical lymphoid cells in the dermis and subcutis (H&E, ×200).

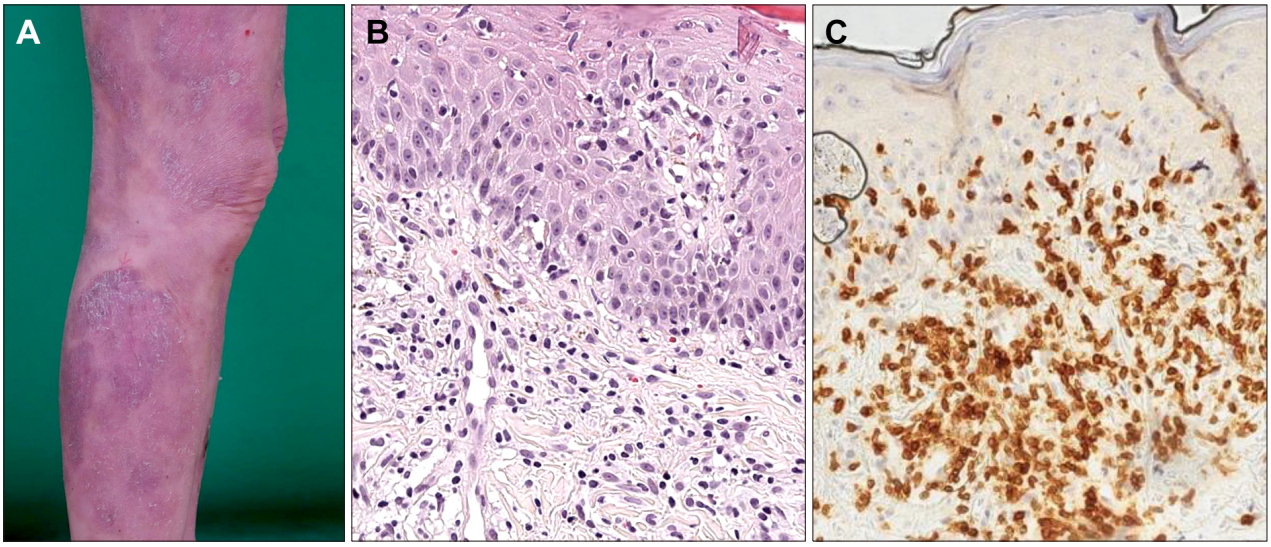

Fig. 2. (A) Erythematous scaly patches on the thigh and the lower leg. (B) Epidermal and dermal infiltration of atypical lymphocyte (H&E, ×200) (C) Immunohistochemistry for T-cell receptor (TCR)-βF1. Biopsy specimen shows TCR-βF1 positivity of lymphocytic infiltrate. (H&E, ×200).

and CD8−. Also immunostaining revealed TCR-βF1+ and TCR-cγM1−. The patient was diagnosed with MF following PCGD-TCL. She received 11 cycles of brentuximab therapy.

The different types of lymphomas coexisting in the same patient are classified into three categories according to the Working Formulation of non-Hodgkins lymphoma[2]. Discordant lymphomas are two histologically distinct lymphomas at two different anatomic sites. Composite lymphomas have two types of lymphoma within the same anatomic lesion. Secondary lymphoma is lymphoma which appeared later when two different lymphomas occur sequentially[2].

PCGD-TCL was introduced as a definitive entity in the 2008 World Health Organization classification of lymphomas[1]. The tumor cells characteristically have a TCR-βF1−, CD3+, CD2+, and CD56+ phenotype and most cases lack both CD4 and CD8[1]. On the other hand, MF shows epidermotropic proliferation of small-to medium-sized T-cells. Mostly, tumor cells have a mature CD3+, CD4+, CD45RO+, and CD8− T-cell phenotype[1].

MF is a risk factor for development of other hematological disorders such as lymphomatoid papulosis, CD30+ anaplastic large cell lymphoma and B-cell lymphoma[3-5]. In addition, several cases were reported that molecular analyses revealed that the same neoplastic clone of T lymphocytes was present in MF and associated lymphoma[3]. In this case, however, different lymphomas with independent findings of immunohistochemistry occurred sequentially, MF occurring after PCGD-TCL. There is no reported case

like our patient in the literature. One possible explanation could be proposed for this association. After receiving treatment of the primary lesion, the condition of patient with or without immunodeficiency is more likely to be vulnerable to secondary malignancy.

In conclusion, we emphasize that when patients diagnosed with cutaneous lymphoma present new skin lesions, clinicians should consider skin biopsies and further immunostaining.

CONFLICTS OF INTEREST

The authors have nothing to disclose.

REFERENCES

1. Sabattini E, Bacci F, Sagramoso C, Pileri SA. WHO classification of tumours of haematopoietic and lymphoid tissues in 2008: an overview. Pathologica 2010;102:83-87.
2. National Cancer Institute sponsored study of classifications of non-Hodgkin's lymphomas: summary and description of a working formulation for clinical usage. The Non-Hodgkin's Lymphoma Pathologic Classification Project. Cancer 1982; 49:2112-2135.
3. Zackheim HS, Jones C, Leboit PE, Kashani-Sabet M, McCalmont TH, Zehnder J. Lymphomatoid papulosis associated with mycosis fungoides: a study of 21 patients including analyses for clonality. J Am Acad Dermatol 2003;49:620-623.
4. Marschalkó M, Csomor J, Eros N, Szigeti A, Hársing J, Szakonyi J, et al. Coexistence of primary cutaneous anaplastic large cell lymphoma and mycosis fungoides in a patient with B-cell chronic lymphocytic leukaemia. Br J Dermatol 2007;157:1291-1293.
5. Barzilai A, Trau H, David M, Feinmesser M, Bergman R, Shpiro D, et al. Mycosis fungoides associated with B-cell malignancies. Br J Dermatol 2006;155:379-386.

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Pigmented Onychomatricoma Showing a Longitudinal Melanonychia: A Case Report and Brief Review of Literature

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Dear Editor:

Onychomatricoma is a rare benign fibroepithelial tumor of the nail unit. It typically presents as thickening of the nail plate, splinter hemorrhage, xanthonychia, and transverse overcurvature, and generally arises in patients with pale skin. Onychomatricoma rarely manifests longitudinal melanonychia (LM). In this case, it is termed pigmented onychomatricoma. However, the occurrence of onycho-

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