A Case of Lymphomatoid Keratosis

Min Jee Choi, M.D., Hei Sung Kim, M.D., Hyung Ok Kim, M.D., Kye Yong Song, M.D.1, Young Min Park, M.D.

Department of Dermatology, Seoul St. Mary’s Hospital, College of Medicine, The Catholic University of Korea, 1Department of Pathology, College of Medicine, Chung-Ang University, Seoul, Korea

Lymphomatoid keratosis (LK) is considered to be a rare variant of cutaneous lymphoid hyperplasia, with epidermotropism. We herein report a case of LK which developed on the abdomen of an elderly Korean woman. A 60-year-old woman presented with a 10-year history of a pruritic, solitary, brown to black plaque on the abdomen. Histopathologically, the specimen showed hyperkeratosis, parakeratosis, acanthosis and Pautrier’s micro-abscess in the epidermis, and a lichenoid infiltration of lymphocytes in the dermis, which expressed both B cell and T cell lineage on the immune-histochemical staining. Based on these clinical and histopathological findings, our case was diagnosed as LK. To our knowledge, this is the first case report of LK in the Korean dermatologic literature. (Ann Dermatol 22(2) 219 ∼ 222, 2010)

-Keywords-
Cutaneous lymphoid hyperplasia, Epidermotropism, Lymphomatoid keratosis

INTRODUCTION

Lymphomatoid keratosis (LK) has been considered to be a variant of uni-lesional mycosis fungoides (MF), as it shows epidermotropism that is characteristic of MF3. With its lichenoid keratotic features, it also has been recognized as a subtype of benign lichenoid keratosis2. Recently, Arai et al.1 proposed that LK is a epidermotropic type of cutaneous lymphoid hyperplasia, where differentiation from MF and benign lichenoid keratosis is possible, by comparing the clinocopathological, immunohistochemical and molecular biological findings. To date, there have been 26 case reports of LK in the English literature, but none in the Korean dermatologic literature. Herein, we report the first case of LK in Korea and review the previous cases.

CASE REPORT

A 60-year-old woman presented with a 10-year history of a pruritic, solitary, well-demarcated, scaly, brown to black plaque on the abdomen, measuring 3.8×1.2 cm in size, which had started as a tiny papule (Fig. 1). Dermoscopic findings revealed numerous brown dots and globules at the periphery (Fig. 2). She had no previous personal or familial history of skin cancer and no other significant cutaneous or medical history was found. There was no history of trauma. A skin biopsy from a local clinic 4 years before showed hyperkeratosis, parakeratosis, acanthosis, papillomatosis and hypergranulosis in the epidermis, which was consistent with seborrheic keratosis. Additio-
Fig. 2. Dermoscopic finding of the lesion showed a globular pattern with brown dots and globules at the periphery.

Fig. 3. Hyperkeratosis, parakeratosis, acanthosis, papillomatosis and hypergranulosis in the epidermis which was consistent with seborrhoeic keratosis (H&E, ×40).

Fig. 4. (A, B) Epidermal hyperplasia and epidermotropism in the epidermis and formation of lymphoid follicle and lichenoid infiltration of lymphocytes in the reticular dermis (H&E stain, ×40, ×100). (C) Epidermotropism with Pautrier’s microabscess in the epidermis (H&E stain, ×400). (D) Lichenoid infiltration of lymphocytes in the reticular dermis (H&E stain, ×400).
Finally, a dense infiltration of lymphocytes in the superficial dermis was observed (Fig. 3).

To confirm the previous diagnosis and rule out malignancy, we performed a second skin biopsy from the same lesion. Histopathological findings showed hyperkeratosis, parakeratosis, acanthosis and epidermotropism with Pautrier’s micro-abscess in the epidermis, a lichenoid infiltration of lymphocytes, the formation of lymphoid follicle and hemorrhage in the reticular dermis (Fig. 4). Solar elastosis was not seen. Immuno-histopathologically, the exocytic lymphocytes in the epidermis and the lymphocytes of the lymphoid follicle in the dermis were positive for CD3, CD4, CD8, CD20, CD30 and CD79a (Fig. 5). Genotypically, rearrangement of TCRγ was partially demonstrated. Based on these clinical and histopathological findings, we made a final diagnosis of LK. Complete excision of the lesion was performed. No local recurrence occurred during the ensuing 3 months.

**DISCUSSION**

Evans et al. first reported a case of uni-lesional MF on the right flank area of a 44-year-old male, where the lesion demonstrated histopathological features of epidermotropism with lichenoid activity. Later on, Kossard suggested that the case reported by Evans et al. should have been categorized as LK, thereby introducing the term LK for the first time in 1997. Thereafter, there have been 23 cases of LK reported in the English literature.

LK clinically presents as an asymptomatic, scaly, erythematous plaque on the face or upper trunk of the middle aged personnel, simulating basal cell carcinoma, actinic keratosis, or seborrheic keratosis. Most cases of LK are idiopathic, however several have been associated with
newly encountered antigens from anthropod bites, stings, tattoo, vaccinations, trauma, injection of foreign substances, pierced ear jewelry, and drugs. Al-Hoqail and Crawford reported cases of LK adjacent to seborrheic keratosis and solar lentigo. Coexistence with these diseases make us suspect that LK might be related to solar damage. In our case, though, solar elastosis was not observed in the dermis. Further evaluation is needed to explain the clear relationship between LK and solar damage. The most pathognomonic pathological finding of LK is epidermotropism. Lymphocytes with tropism for the epidermis are composed of both B cells and T cells. Special attention is needed to differentiate LK from unilesional MF because these 2 diseases largely resemble one another. Unilesional MF is a rare variant of MF which usually appears as a single, isolated lesion comprising of less than 5% of the body's surface area. The disease is usually benign and has an excellent response to locally ablative treatments. Uni-lesional MF is different from LK in that the epidermotropic lymphocytes in unilesional MF mainly consist of helper T cells possessing T cell clonality, which conveys the malignant transformation. Other major differentials to consider are lichenoid actinic keratosis and benign lichenoid keratosis. Both of them commonly show lichenoid infiltrates of lymphocytes with epidermal hyperplasia. However, lichenoid actinic keratosis differs from LK in that atypical basal cells and keratinocytes can be found whereas epidermotropism of the lymphocytes are not seen. Benign lichenoid keratosis shows necrotic keratinocytes, spongiosis and Max-Joseph spaces without epidermotropism. Also, irritated seborrheic keratosis and inflamed seborrheic keratosis should be included in the differential diagnosis of benign keratotic dermatosis. Based on the histopathological and immunohistochemical findings, our case was diagnosed as LK. The unusual feature in our case is the history of a transformation from seborrheic keratosis to LK. Either the transformation of seborrheic keratosis to LK is incidental or else these two diseases may be truly related. Chronic rubbing may induce hyperkeratotic changes in the epidermis and exocytosis and spongiosis of lymphocytes in the dermis, reflecting the unidentified culprit factor of LK like other spongiotic dermatitis. For a clear explanation, further investigation is needed. In conclusion, we experienced a typical case of LK that developed on the abdomen and was transformed from seborrheic keratosis. LK is an epidermotropic kind of cutaneous lymphoid hyperplasia, with hyperkeratotic changes in the epidermis, therefore it should be included in the differential diagnosis of benign keratotic dermatosis.

REFERENCES