

A Case of Lichen Spinulosus with an Histologic Finding of Follicular Mucinosis

Dae-Heon Oh, M.D., Kyoung-Tae Park, M.D., Jeong-Soo Kim, M.D., Hee-Joon Yu, M.D.

Department of Dermatology, Hanyang University College of Medicine, Seoul, Korea

Follicular mucinosis is an uncommon inflammatory dermatosis characterized histologically by mucinous degeneration of the outer root sheath of the hair follicle and sebaceous gland. In recent years, follicular mucinosis has been recognized as an incidental histologic reaction pattern seen in association with a variety of neoplasms and dermatoses. We report a case of lichen spinulosus with an incidental histologic finding of follicular mucinosis in a 7-year-old boy. He presented with lichen spinulosus and grouped follicular keratotic papules on the elbows and knees. A biopsy specimen showed findings of both lichen spinulosus and follicular mucinosis, including keratotic follicular plugging, perifollicular lymphocytic infiltration, reticular degeneration, cystic space formations, and abundant mucin deposits in the pilosebaceous follicles. (*Ann Dermatol* 17(2) 79~82, 2005)

Key Words: Follicular mucinosis, Lichen spinulosus

INTRODUCTION

Follicular mucinosis is a general term used to describe a reactive disease process characterized histologically by mucin depositions in hair follicles and sebaceous glands which are undergoing epithelial, reticular degeneration¹. Clinically, the lesions appear as minimally scaling, erythematous plaques with prominent follicles. On the hair-bearing skin of the scalp or face, alopecia is a prominent feature². This process may occur as a primary disorder or it may be associated with benign (e.g., angiolymphoid hyperplasia) or malignant (e.g., mycosis fungoides) diseases³. In our case, follicular mucinosis was associated with lichen spinulosus. Lichen spinulosus is a benign dermatoses characterized by follicular keratotic papules that are grouped into large patches, which may symmetrically involve the elbows, knees, or buttocks¹⁴.

CASE REPORT

A 7-year-old boy presented with a 3-month history of skin-colored follicular keratotic papules. Each individual lesion was a flat to conical projection, 1 to 3 mm in diameter, which bore a hair-like, horny spine, which protruded approximately 1 to 2 mm above the surface. The lesions were distributed symmetrically on both elbows and knees. He had no subjective symptoms such as paresthesia or pruritus. His past and family history was unremarkable. On physical examination, there were no abnormal findings other than the cutaneous lesions. Laboratory investigations, including a complete blood count, erythrocyte sedimentation rate, liver and renal function tests and urinalysis were all within normal limits. Chest roentgenograms were unremarkable. Clinically, the patient was diagnosed with lichen spinulosus.

A biopsy specimen taken from the right elbow showed a compact, keratinous plug in the dilated hair follicle, with perifollicular mononuclear cell infiltrates. In the pilosebaceous follicle, there were reticular degenerations, cystic space formations, and abundant mucin deposits in the cystic space and outer root sheath. Mucin deposits stained metachromatically with Giemsa, toluidine blue, and alcian blue in acid PH. Histopathologically, a biopsy

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Reprint request to: Jeong-Soo Kim, M.D., Department of Dermatology, Hanyang University Kuri Hospital, 249-1 Gyomun-dong, Kuri-city, Kyunggi-do 471-701, Korea.
Tel: 82-31-560-2280, Fax: 82-31-557-4872
E-mail: Tuentuen@hanyang.ac.kr

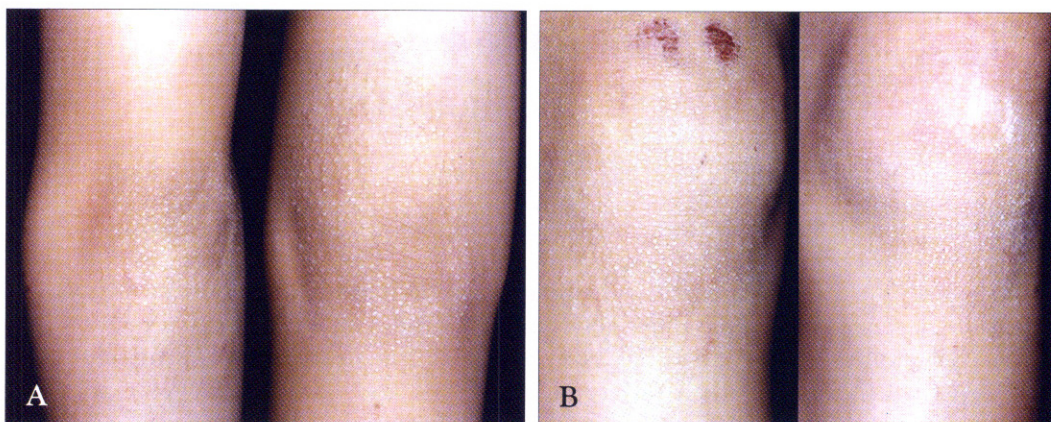


Fig. 1. Skin-colored, grouped keratotic papules on both elbows (A) and knees (B).



Fig. 2. Closed lesions showing a hair-like, horny spine on the right knee.

specimen showed findings of both lichen spinulosus and follicular mucinosis.

The patient was treated with topical adapalene and methylprednisolone aceponate. There was clinical improvement within one month.

DISCUSSION

Pinkus⁵ described alopecia mucinosa as an inflammatory condition characterized by mucinous follicular degeneration in well-demarcated plaques associated with alopecia. As alopecia may not be grossly evident in areas other than the scalp and beard, the term follicular mucinosis is preferred². Braun-Falco⁶ recognized the two forms of Follicular mucinosis, one occurring as a benign condition and the other associated with lymphoma. The cause is unknown,

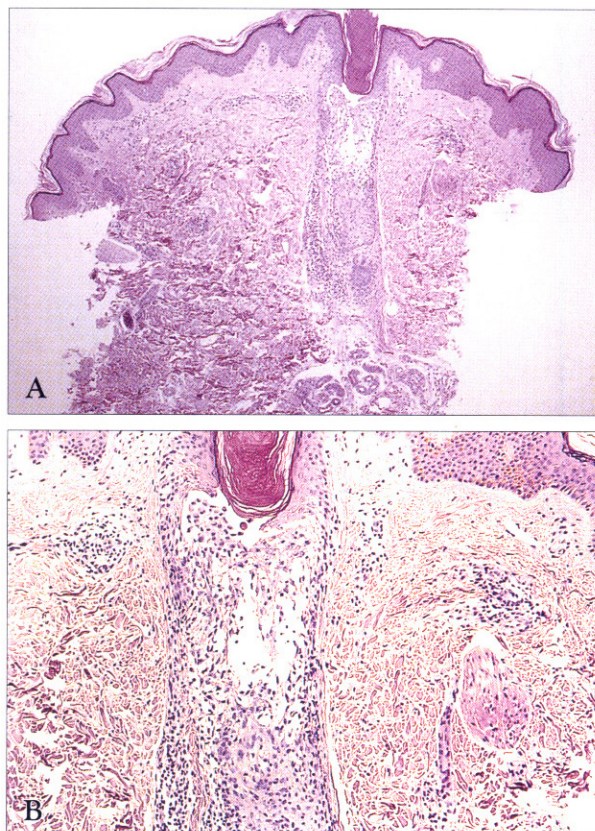


Fig. 3. A low-power view showing a keratinous plug in the dilated hair follicle, perifollicular mononuclear cell infiltrates, reticular degeneration, cystic space formations, and abundant mucin deposits in the pilosebaceous follicle (A: H&E, $\times 100$, B: H&E, $\times 200$).

but immunopathologic studies have indicated that altered cell-mediated immune mechanisms may play

a role in its pathogenesis. It has been suggested that cytokines released from T-lymphocytes may stimulate the epithelium to secrete mucin^{1,3}. Clinically, it most commonly presents as erythematous papules or plaques, with follicular prominence and scaling. There may only be one lesion, especially on the head or neck. Multiple lesions may also occur. Lesions are distributed most commonly on the face, neck, and scalp but may appear on any part of the body^{2,4,7}. Our patient presented with grouped follicular keratotic papules on both his elbows and knees, and the clinical findings indicated the condition to be lichen spinulosus.

Emmerson⁷ further classified patients with follicular mucinosis into three groups. In the first group, lesions were usually limited to the face and cleared spontaneously within a few months. In the second group, lesions were either confined to the face or were widespread and pursued a chronic and relapsing course over several years. In both these groups, there was no underlying disease, and patients were generally younger than 40 years. The third group comprised of patients with follicular mucinosis, in association with lymphoma, and predominately included older adults with widespread lesions. In more recent years, follicular mucinosis has been recognized as an incidental histologic reaction pattern seen in association with a variety of neoplasms and dermatoses including angiolymphoid hyperplasia⁸, lupus erythematosus⁹, familial reticuloendotheliosis¹⁰, Goodpasture syndrome¹¹, melanocytic nevi¹², and lentigo maligna¹³. We describe a case of lichen spinulosus with an incidental histologic finding of follicular mucinosis. As stated above, we believe that follicular mucinosis is associated with lichen spinulosus in a nonspecific reaction pattern. In the literature review, follicular mucinosis has not been previously reported in association with lichen spinulosus.

Lichen spinulosus was first described by Crocker in 1883 as a benign follicular eruption seen primarily in the younger generation, and characterized by follicular keratotic papules involving the elbows, knees, and buttocks¹⁴. The individual lesion is a flat to conical projection, 1 to 3 mm in diameter, that bears a hair-like, horny spine. This spine protrudes approximately 1 to 2 mm above the surface. The cause is not currently known, but there is a strong possibility of toxic agents, vitamin A deficiency, trauma, infections, or drugs being involved. Some have proposed that lichen spinulosus is a type of

follicular nummular eczema¹⁵.

There is no effective standard treatment for follicular mucinosis. Topical and intralesional corticosteroids have produced varying degrees of improvement. Dapsone, PUVA, radiation therapy, interferon alfa-2b, mepacrine, and indomethacin have been reported to be effective in individual cases^{2,4}. The predominant treatment for lichen spinulosus has been mild keratolytics such as 3% resorcin or salicylic acid ointment. Ammonium lactate and tretinoin are other alternatives^{14,15}. For treatment of both follicular mucinosis and lichen spinulosus, our patient was treated with topical adapalene and corticosteroid and improvement was seen within one month.

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