

CASE REPORT

Angiolymphoid Hyperplasia with Eosinophilia on the Palm

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Angiolymphoid hyperplasia with eosinophilia (ALHE) is an uncommon dermal angioproliferating tumor, characterized by red to brown papules or nodules on the head and neck, though also occurring in the mouth, trunk, extremities and inguinal area. The palm is a very unusual site for ALHE, and there have been very few cases reported globally thus far. ALHE can be pruritic and painful and histopathologic findings show vascular proliferation with infiltration of eosinophils and lymphocytes in the dermis. Plump endothelial cells protrude into the lumen. We report a case of ALHE occurring at an unusual site, the right palm, in a 62-year-old man, who had suffered from a solitary pinkish-colored, central depressed round hyperkeratotic plaque on his palm for 4 years. On the basis of clinical and histopathologic data, a diagnosis of ALHE was made. To our knowledge, this is the first report of ALHE on the palm in Korean dermatologic literature. (*Ann Dermatol* 22(3) 358 ~ 361, 2010)

-Keywords-

Angiolymphoid hyperplasia with eosinophilia, Epithelioid hemangioma, Palm

INTRODUCTION

Angiolymphoid hyperplasia with eosinophilia (ALHE) usually presents as red- to brown-colored papules or

nodules on the head and neck, especially on the ears and scalp. It also occurs on the mouth, trunk, extremities and inguinal area. It mainly affects middle-aged woman, can be pruritic or painful, and can present as solitary or multiple lesions¹. The etiology of ALHE is unknown and it must be carefully differentiated from Kimura's disease which shares similar clinicohistopathological features^{2,3}. Our patient presented with a solitary pinkish-colored pruritic plaque on the right palm. We herein report a case of ALHE occurring at the unusual site of the palm.

CASE REPORT

A 62-year-old Korean man presented with a solitary, pinkish-colored, central depressed round hypertrophic plaque on the right palm (Fig. 1). He first noticed the lesion 4 years previously and had it excised 2 years ago only for it to recur at the same site. On his second visit to us, he reported mild pruritis of the lesion but no other medical problems. Physical examination revealed a solitary pinkish-colored, 15-mm sized, central depressed,



Fig. 1. Solitary, pinkish-colored, central depressed, round hyperkeratotic plaque on the right palm.

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round hyperkeratotic plaque on the right palm. There were no palpable regional or systemic lymph nodes. Excisional biopsy and routine laboratory testing including complete blood count, blood chemistry, urine analysis were performed. All laboratory tests were within normal

limits and there was no peripheral eosinophilia. Histopathologic examination of the first biopsy showed vascular proliferation and dilation in the dermis, and perivascular infiltration of the inflammatory cells. Examination of the second biopsy revealed similar findings:

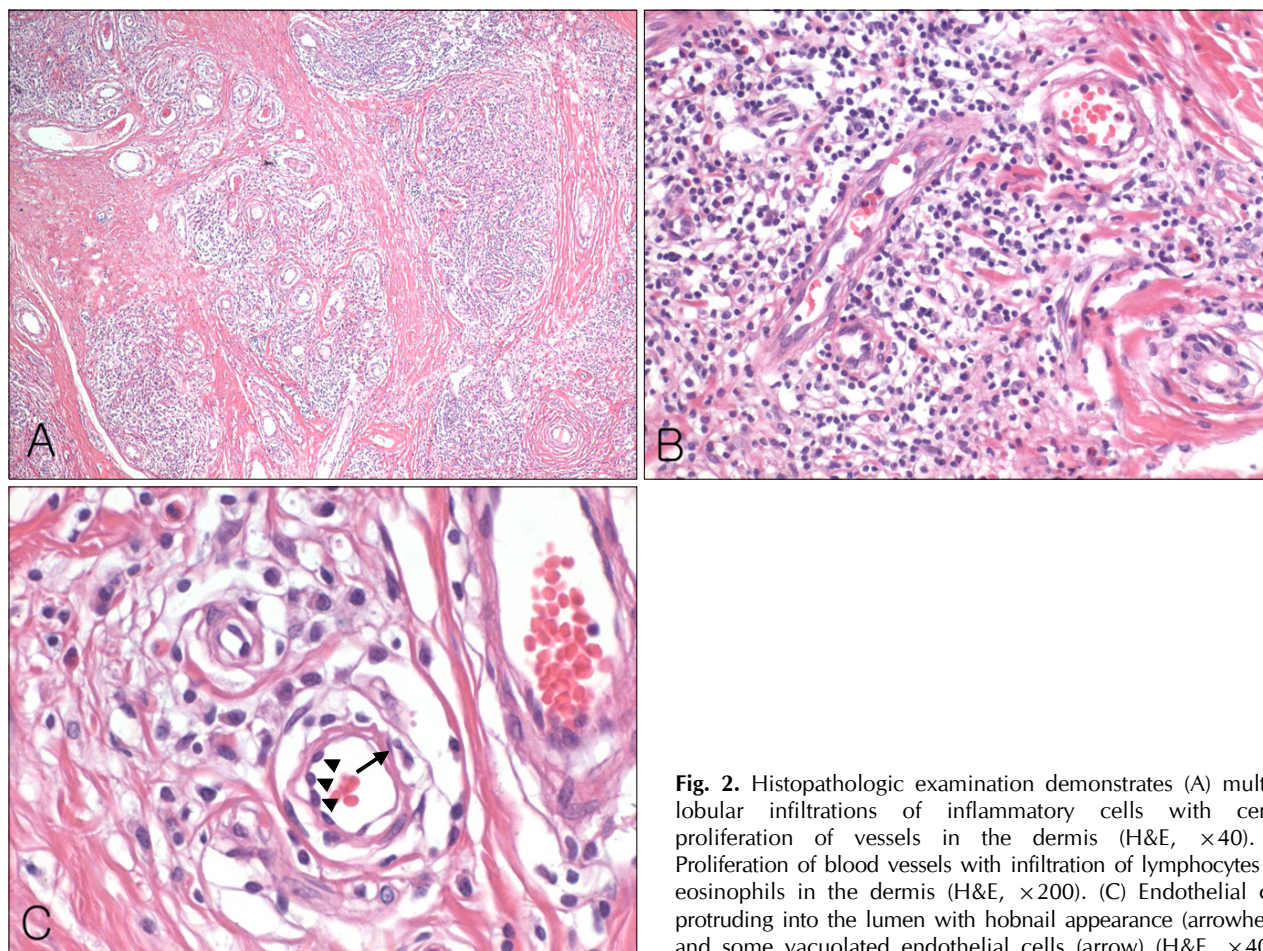


Fig. 2. Histopathologic examination demonstrates (A) multiple lobular infiltrations of inflammatory cells with central proliferation of vessels in the dermis (H&E, $\times 40$). (B) Proliferation of blood vessels with infiltration of lymphocytes and eosinophils in the dermis (H&E, $\times 200$). (C) Endothelial cells protruding into the lumen with hobnail appearance (arrowheads) and some vacuolated endothelial cells (arrow) (H&E, $\times 400$).

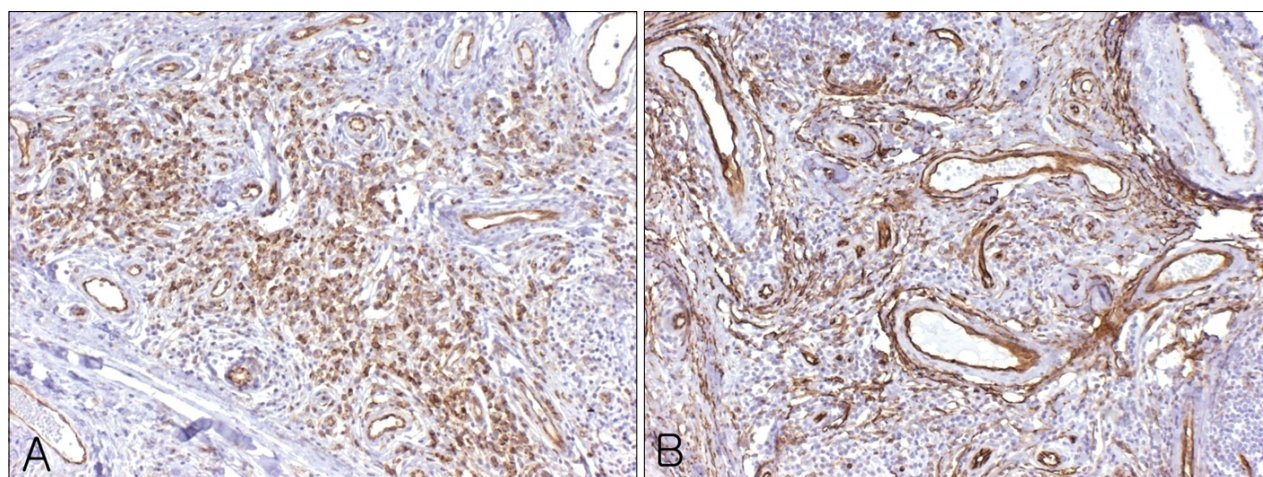


Fig. 3. Endothelial cells are positive in (A) CD31 ($\times 100$) and (B) CD34 ($\times 100$).

much vascular proliferation with lymphocyte and eosinophil infiltration in the dermis. Plump endothelial cells protruding into the vessel lumen and vacuolated endothelial cells were seen. There was no lymphoid follicle formation (Fig. 2). Immunohistochemistry showed that the endothelial cells were positive for CD34 and CD31 (Fig. 3). The patient was diagnosed with ALHE, and treated with total excision. Although the tumor recurred once because of incomplete excision, there was no recurrence during 22 months of follow-up period after a more radical second excision.

DISCUSSION

ALHE is an uncommon cutaneous angioproliferative disorder manifested by solitary or multiple, superficial or subcutaneous, red to brown firm papules and nodules. It was first described by Wells and Whimster¹ in 1969 and though its pathogenesis remains unclear it is thought to be associated with inflammation, infection, and arteriovenous malformation⁴. ALHE is also known as epithelioid hemangioma⁵. The lesions are often associated with pruritus, pain, and spontaneous bleeding, and may coalesce into confluent plaques. ALHE can occur in all races, but is reported more frequently in Asians. Women are more commonly affected than men, and the disease is more prevalent in young to middle-aged adults than in the children or elderly^{6,7}. The head and neck are characteristically affected, but the disease is also known to affect the trunk, extremities and inguinal area. The hands, and particularly, the palms, are an unusual location for ALHE⁸, with very few reported cases of ALHE on the palms. There are very few reported cases worldwide of ALHE on the palm, and no such previous reports in the Korean demographic literature. Regional lymphadenopathy (5~20%) or peripheral eosinophilia (20%) are present at low frequencies⁹. In our case, there was no regional or systemic lymphadenopathy and no peripheral eosinophilia.

Histopathologically, there is proliferation of large endothelial cells lining vascular spaces, and lymphocytic and eosinophilic inflammatory infiltrations in the dermis. Plump endothelial cells show scalloped borders, lobulated nuclei, and mitoses. The "epithelioid" endothelial cells which protrude into the vascular lumen, have "cobblestone" appearance. Vacuolated endothelial cells can also be seen⁹.

The most important differential diagnosis of ALHE is Kimura's disease. Clinically, Kimura's disease occurs exclusively in young oriental males, presenting with deep, large subcutaneous nodules which are often solitary.

Systemic lymphadenopathy (70%) and marked peripheral eosinophilia are very common. Histopathologically, well-developed lymphoid follicles, eosinophilic abscess formations and a lack of change in vessels are characteristic of Kimura's disease. ALHE, in contrast, presents with more superficial lesions, no lymphoid follicle formation and typically no systemic lymphadenopathy or peripheral eosinophilia. Furthermore, increased serum IgE levels, concomitant asthma, proteinuria, and nephrotic syndrome, which are common in Kimura's disease, are not common in ALHE⁹.

Histopathologically, our case revealed vascular proliferation with infiltration of eosinophils and lymphocytes in the dermis. As in typical ALHE, plump endothelial cells protruded into the lumen and vacuolated endothelial cells were seen in the vascular lumen. Lymphoid follicles were not seen (Fig. 2). There were no clinical manifestations such as palpable lymph nodes, laboratory abnormalities, and peripheral eosinophilia. On the basis of these findings, we diagnose the lesion as ALHE.

While surgical excision is regarded as the treatment of choice for ALHE, spontaneous regression is a known possibility¹⁰. Alternative treatments include topical or intralesional corticosteroid, lasers, electrodesiccation, radiation, and cryotherapy, though recurrences have also been reported with these, particularly after incomplete removal¹¹. In our case, the lesion recurred after the first excision. Since the recurrence seemed to be due to incomplete excision of the mass, the second excision was performed more radically and the mass was removed completely. There was no recurrence after 22 months follow-up.

In the Korean dermatologic literature, 17 cases of ALHE have been reported thus far. The scalp, ears, and legs were commonly affected, with the arms, trunk, forehead, lower eyelids, and fingers also occasionally involved. To our knowledge, this is the first report of ALHE occurring on the palm in Korean dermatologic literature.

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