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CASE REPORT

A Case of Eccrine Angiomatous Hamartoma Associated with Verrucous Hemangioma

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Eccrine angiomatous hamartomas are benign vascular and eccrine malformations often accompanied by hyperhidrosis or pain, increased eccrine glands, and aggregates of vessels. Verrucous hemangiomas are congenital vascular malformations presenting as unilateral grouped papules. Histologically, they show verrucous epidermal change and proliferation of capillaries in the dermis. We report a case of a 47-year-old woman with a red keratotic patch overlying a bluish plaque on the right sole, which had been present since birth. It was accompanied by pain and hyperhidrosis. Histologically, there were verrucous changes in the epidermis, numerous dilated capillaries in the papillary dermis, and increased eccrine glands with angiomatous foci in the deep dermis. The epithelial cells of the eccrine glands were positive for CEA, and the endothelial cells were positive for CD31 and GLUT-1. Eccrine angiomatous hamartomas have been reported in conjunction with other vascular tumors in only a few instances. We report an interesting case of an eccrine angiomatous hamartoma associated with a verrucous hemangioma. **(Ann Dermatol 21(3) 304～307, 2009)**

-Keywords-
Eccrine angiomatous hamartoma, Verrucous hemangioma

INTRODUCTION

Eccrine angiomatous hamartomas (EAH) are benign cutaneous hamartomas, often localized in the distal extremities of children or young adults. They are usually asymptomatic, although they may be painful and hyperhidrotic[1]. EAHs are composed of enlarged eccrine sweat glands intimately associated with small blood vessels located in the middle to deep dermis. Five cases of EAH have been reported in the Korean literature. However, EAH associated with verrucous hemangioma has not been reported in the Korean literature. We report an interesting case of EAH associated with verrucous hemangioma.

CASE REPORT

A 47-year-old Korean woman presented to a dermatology clinic with a skin lesion on the right sole, which had been present since birth. It had been increasing in size over a 1-month period after the patient had sustained trauma. The patient also noted pain and focal hyperhidrosis. Her personal and family histories were unremarkable. Physical examination revealed an erythematous keratotic plaque overlying a 5-cm sized, bluish, ill-defined plaque on the right sole (Fig. 1).

An incisional biopsy showed that the lesion had marked

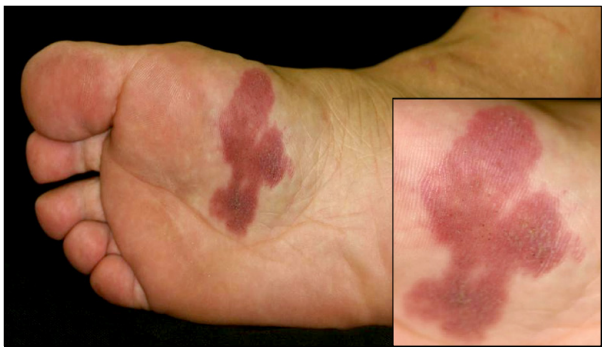

Fig. 1. An erythematous keratotic plaque overlying a bluish, ill-defined plaque on the right sole.

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verrucous changes, including hyperkeratosis, papillomatosis, and irregular acanthosis in the epidermis, numerous dilated capillaries, and some congested vessels in the papillary dermis (Fig. 2, 3A). A nodular proliferation of eccrine glands was intimately admixed with numerous small vessels in the middle to deep dermis (Fig. 2, 3B). The epithelial cells of the eccrine glands were positive for CEA (Fig. 4), and the endothelial cells of numerous dilated capillaries were positive for CD31 and GLUT-1 (Fig. 5).
On the basis of these histological findings, we made a diagnosis of EAH associated with verrucous hemangioma.

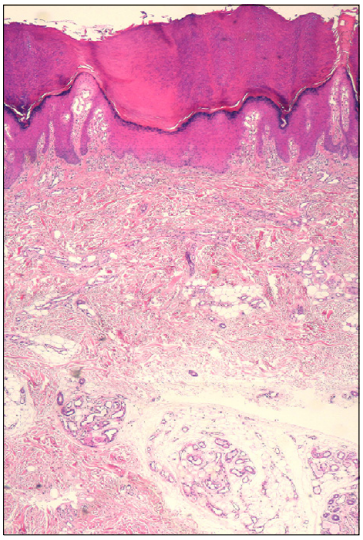

Fig. 2. Epidermal hyperplasia, an increased number of vessels in the superficial dermis, and eccrine structures with intermingled vascular channels in the deep dermis (H&E, ×20).

DISCUSSION

EAHs are vascular and eccrine malformations, often localized in the distal extremities. They may be congenital or may appear later in childhood[1]. Clinically, they present as solitary papules or, rarely, in multiple distribution. They may be red, yellow, or brown in color. They are usually asymptomatic, although they may be painful and hyperhidrotic. Histologically, EAHs show enlarged eccrine sweat glands in the middle and lower dermis, intimately associated with thin-walled, aggregated vessels. The secretory portions of the eccrine glands are positive for S-100, CEA, and EMA.
EAHs have been reported in combination with other vascular tumors in a few cases. One had elements of an arterio-venous malformation, in which vessels showed uneven proliferation of intima and muscular walls[2]. Another was associated with a spindle cell hemangioma[3]. It contained vascular components consisting of dilated vessels intermixed with spindle cells[3]. A case of EAH with overlying verrucous hemangioma-like features has been described. However, the vessels were negative for GLUT-1, supporting the impression of hamartoma[4].
Epidermal proliferation with verrucous features has been found in rare cases of EAH[5,6]. However, in these cases, other findings of verrucous hemangiomas, such as dilated capillaries and congested vessels in the papillary dermis, have not been observed.
Verrucous hemangiomas are rare vascular malformations, first described by Imperial and Helwig[7] in 1967. They are a structural variant of capillary or cavernous hemangiomas, in which reactive epidermal changes develop secondarily. They usually involve the lower

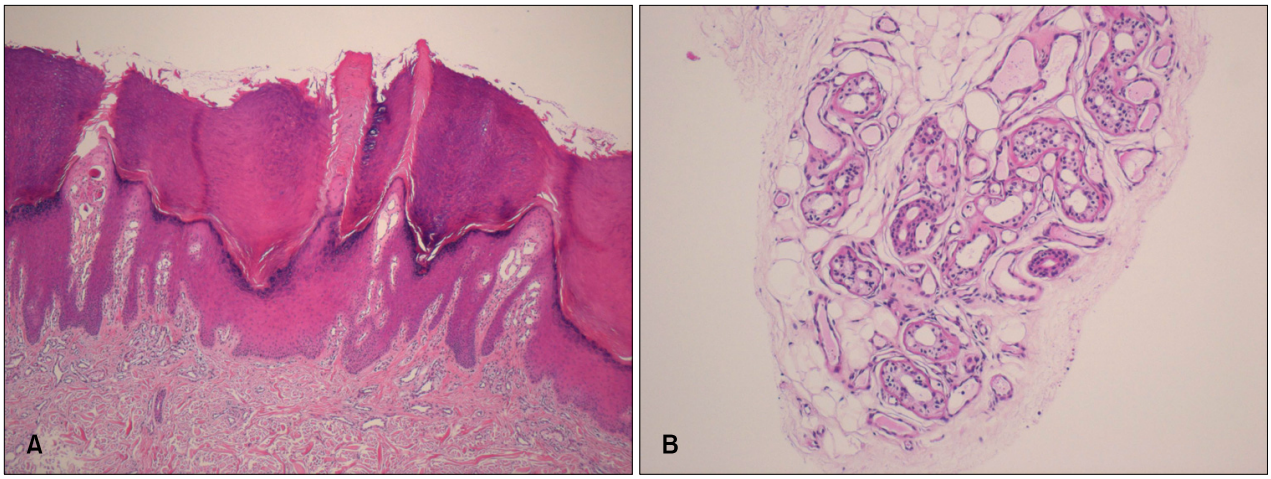

Fig. 3. (A) Verrucous changes in the epidermis and numerous dilated capillaries in the papillary dermis (H&E, x40). (B) Increased eccrine glands with angiomatous foci in the deep dermis (H&E, ×100).

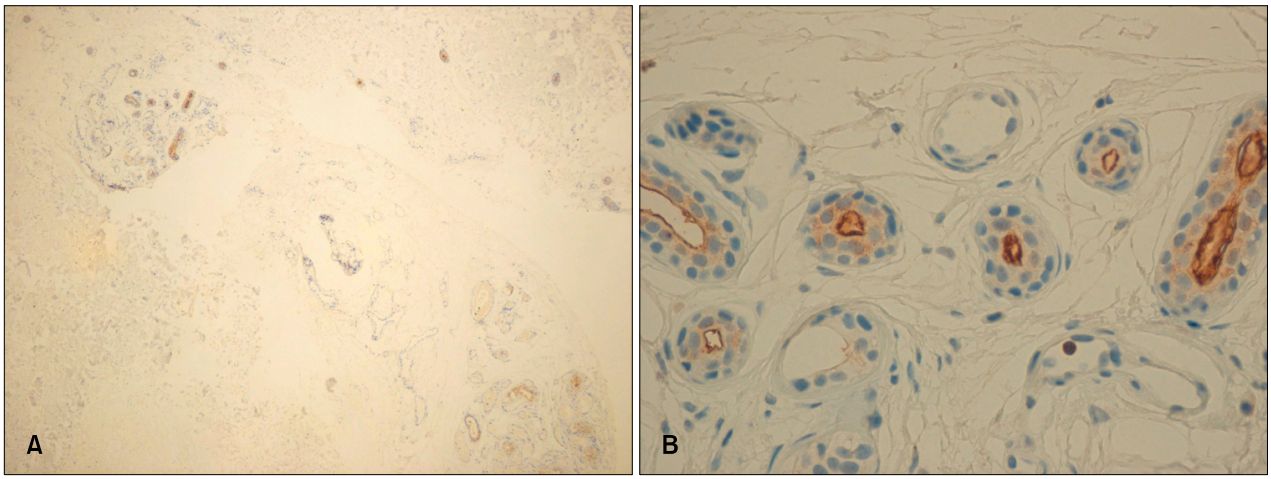

Fig. 4. Epithelial cells in the eccrine glands are positive for CEA (A: ×40, B: ×400).

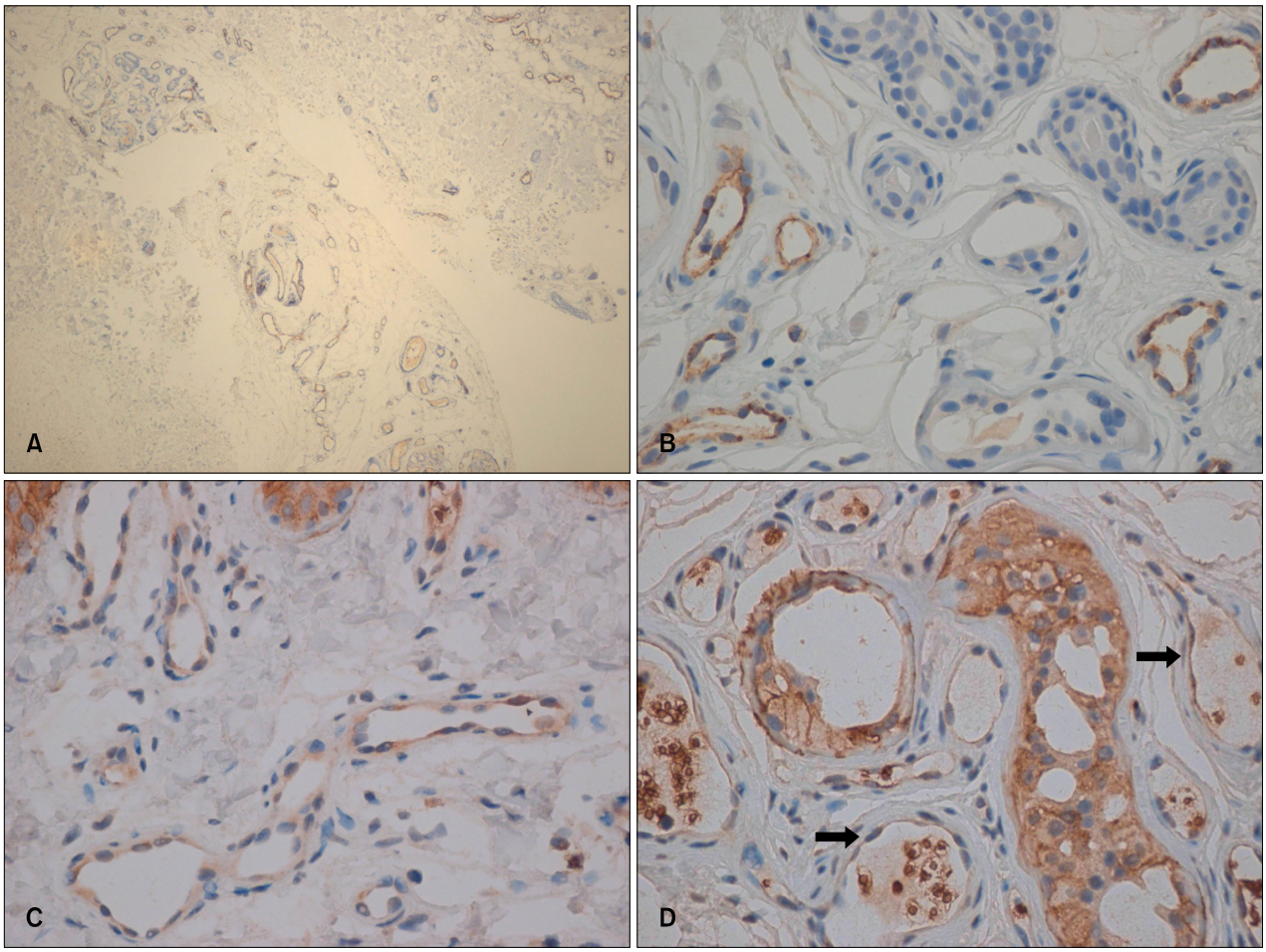

Fig. 5. Endothelial cells in the vessel wall are positive for CD31 and GLUT-1 (A: CD31, ×40, B: CD31, ×400, C, D: GLUT-1, ×400).

extremities, and their onset is near the time of birth or, less frequently, in early infancy[7]. The clinical appearance of a verrucous hemangioma changes with time[7]. Initially, it is a unilateral and circumscribed grouped of warty

papules or plaques, bluish red in color. Later, it develops into a hyperkeratotic hemangiomatous papule in a linear or serpiginous configuration. Finally, the lesion becomes verrucous as a result of trauma and secondary infection. Histologically, verrucous hemangiomas appear initially as capillary hemangiomas concentrated in the dermis, and they adopt a verruciform pattern with papillomatosis, irregular acanthosis, and cavernous or mixed hemangiomas in the dermis and subcutaneous tissue[8]. Immunostaining shows focal glucose transporter-1 protein (GLUT-1) endothelial positivity[9].

Our case showed marked verrucous changes in the epidermis and numerous dilated capillaries in the papillary dermis. There was positive GLUT-1 immunostaining in the endothelial cells of numerous dilated capillaries. According to these findings, a diagnosis of verrucous hemangioma was favored over hamartomatous change or angiokeratoma circumscriptum, which is an acquired dermatosis histologically characterized by dilatation of the vessels of the papillary dermis, without involvement of the deep dermis or hypodermis, and with hyperkeratosis[10].

To our knowledge, no investigators have reported the co-existence of EAH and verrucous hemangioma with GLUT-1 positivity. Therefore, we report an interesting case in which these two entities co-exist.

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