Eccrine Angiomatous Hamartoma in a Patient with Nevus Depigmentosus and Nevus Spilus

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Sir,

Eccrine angiomatous hamartoma (EAH) is a relatively rare hamartoma of eccrine glands and blood vessels in infancy. Clinically, EAH usually presents with a solitary nodule or plaque, involving the distal extremities.

A 3-year-old boy visited the Department of Plastic Surgery of our hospital, complaining of a painful nodule on the hand which had existed since birth. Physical examination revealed a skin-colored, smooth, small nodule on the left thumb [Figure 1a]. Neither palmar hyperhidrosis nor finger hyperplasia was observed. In addition, the patient had a depigmented macule on the left abdomen [Figure 1b] and a pigmented freckle on the right upper extremity [Figure 1c], both of which were developed soon after birth. All the skin lesions were considered benign; however, according to the parents’ wish, the digital nodule was completely resected and biopsy was performed from the depigmented macule under general anesthesia. Histological examination showed nodular clusters composed of eccrine glands and dilated vessels in the lower dermis [Figure 2a]. Immunohistochemistry showed a number of CD31-positive vessels within the foci of eccrine sweat glands [Figure 2b]. A biopsy specimen from the depigmented macule showed no specific features. Unfortunately, histological examination was not carried out on the nevus spilus on the arm. The patient has been followed up without local recurrence.

EAH usually presents with a papule, nodule, plaque, patch, or macule.[1-4] Verrucous lesions, hemangioma-like lesions, and verrucous hemangioma-like lesions are rarely reported.[5-7] The diagnostic criteria are hyperplasia of normal or dilated eccrine glands, close association of the eccrine structures with capillary angiomatous foci, and the presence of pilar, lipomatous, mucinous, or lymphatic structures.[4] EAH is often accompanied by local pain, hypertrichosis, and local hyperhidrosis. Pelle et al.[8] reviewed 37 cases of EAH and described that pain/tenderness was observed in 42.4% and sweating in 34.3%.

EAH is a hamartoma and has been reported to occur in a patient with neurofibromatosis Type I[9] and in one with Cowden syndrome.[10] Our case developed other congenital nevi such as nevus depigmentosus and nevus spilus. We examined throughout the body, but did not detect congenital hamartomas or nevoid lesions of any other origins. Unfortunately, because the patient was an infant, detail examinations such as sweating test, echography, or magnetic resonance imaging were not carried out. Although association of nevus depigmentosus and nevus spilus has rarely been reported,[11] cooccurrence of EAH with either nevus depigmentosus or nevus spilus has not been reported to date. Therefore, the etiopathogenesis of the rare cooccurrence of the three conditions is still unknown. Although the coexistence may be only incidental, accumulation of similar cases is needed in the future. The prognosis of EAH is good, and thus, surgical excision is occasionally chosen, especially in cases accompanying local pain.

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Nil.

Conflicts of interest
There are no conflicts of interest.

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