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Congenital Linear Smooth Muscle Hamartoma with Hypertrichosis: Hair Density on Dermoscopy in Parallel with the Number of Smooth Muscle Bundles

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Dear Editor:

Smooth muscle hamartoma (SMH) is a rare benign tumor of the skin, characterized by the proliferation of smooth muscle bundles within the reticular dermis^{1,2}.

A 5-month-old girl presented with multiple skin-colored patches with hypertrichosis on the left upper back since birth (Fig. 1A). The lesions were composed of three patches with linear distribution (Fig. 1B). The mother of the infant had noted transient induration with piloerection of the lesions when exposed to cool air and rubbing, called a "pseudo-Darier sign." Dermoscopic findings showed different types of hypertrichosis in the three patches (Fig. 1D~F). Histopathological examination revealed numerous haphazardly arranged smooth muscle bundles in the dermis (Fig. 1C), and immunohistochemical staining showed diffusely stained smooth muscle actin (Fig. 1G~I). These findings suggested a diagnosis of congenital SMH.

Gagné and Su³ suggested that hypertrichosis or prominent overlying hair in congenital SMH was usually present, but hair density was unchanged besides increased hair diame-

ter and length. However, we speculated that there is a relation between hypertrichosis including hair density and the amount of smooth muscle bundle. Interestingly, dermoscopy revealed hypertrichosis with varying densities at the different sites (Fig. 1D~F), and histopathological examination revealed different numbers of smooth muscle in the reticular dermis (Fig. 1G~I). Thus, we measured the density of the hair (/mm³) in each lesion by using dermoscopy and the area of smooth muscle in the reticular dermis (%) by using a digital image analysis software (ImageJ 1.01 version; National Institutes of Health, Bethesda, MD, USA). The density of hair was 0.27/mm³ in the lateral hairy patch (Fig. 1D), 0.44/mm³ in the middle patch (Fig. 1E), and 1.40/mm³ in the medial patch (Fig. 1F), and the number of smooth muscle bundles in the reticular dermis was 9.5%, 24.6%, and 31.0% in each patch (Fig. 1G~I). Therefore, we believed that the number of hair in multiple patches, as observed using dermoscopy, was in proportion with the number of smooth muscle bundles in each lesion, as observed in the histopathological examination

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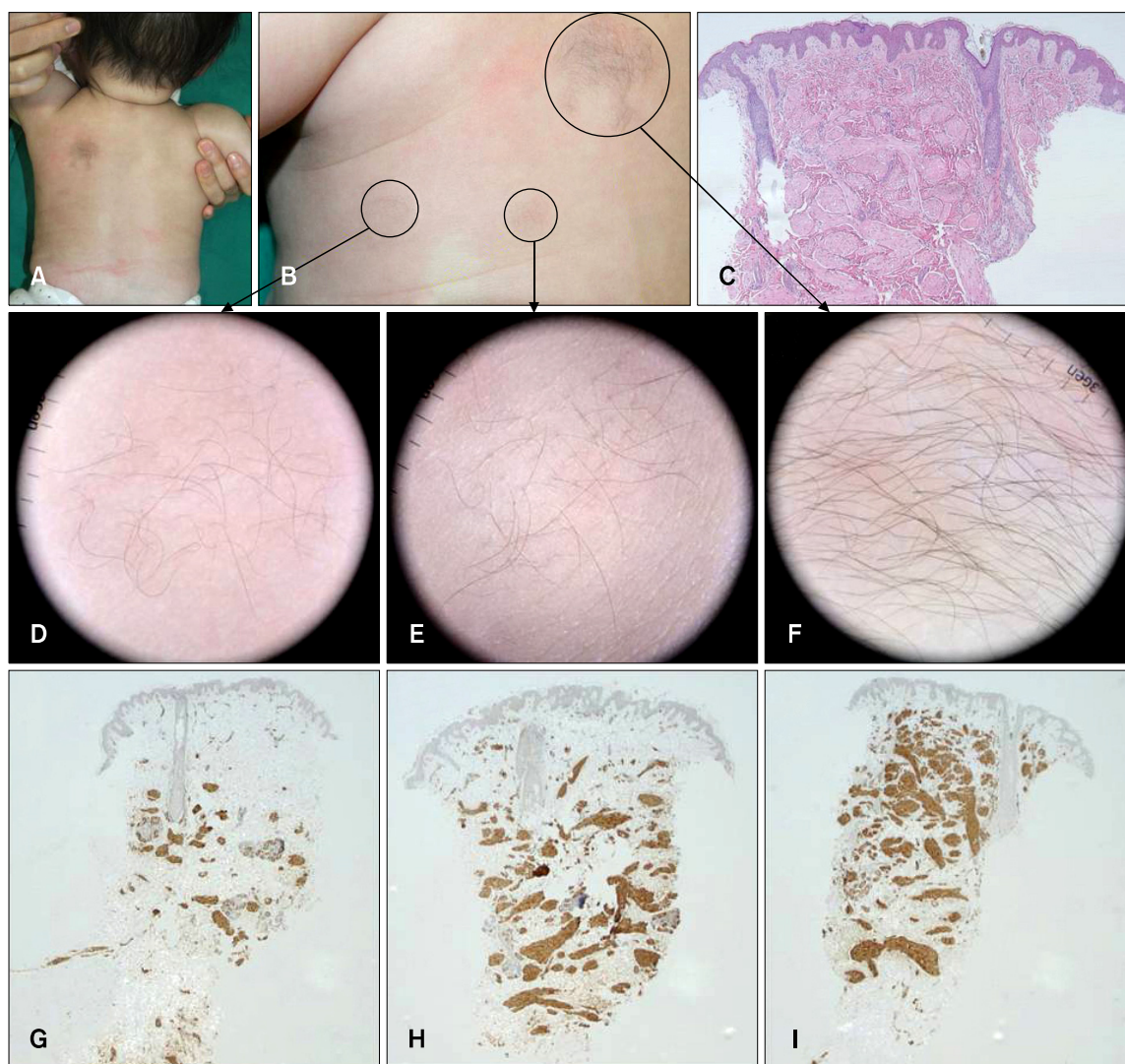


Fig. 1. (A) Multiple linear scattered skin-colored patches with hypertrichosis on left upper back were observed. (B) Close-up view of left upper back. (C) Numerous smooth muscle bundles with various direction throughout dermis were shown (H&E, $\times 40$). (D~F) In three sites, hypertrichosis with various density was shown on dermoscopy. (G~I) Different amount of smooth muscles according to hypertrichosis was observed in the reticular dermis (smooth muscle actin, $\times 20$).

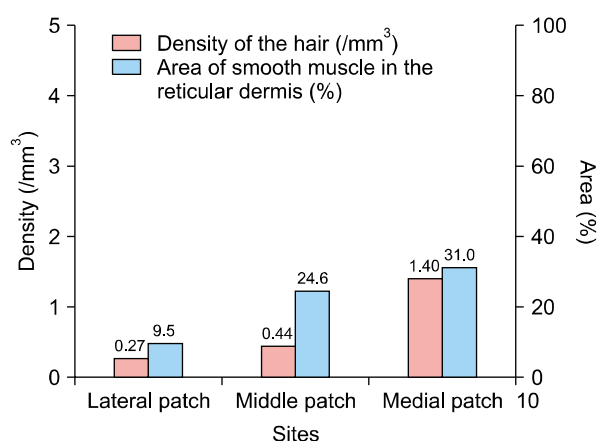


Fig. 2. Correlation between hypertrichosis and the amount of smooth muscle bundle.

(Fig. 2).

In the literature, SMH probably represents aberrant development of the arrector pilorum during fetal maturation. In human skin, it may develop at the time of mesodermal maturation⁴. Koizumi et al.⁵ suggested that numerous CD34-positive cells may be present in the stroma surrounding the smooth muscle bundles. It has been speculated that dermal dendritic cells release growth factors or directly contact the epithelial cells of the bulge, a region considered to represent a reservoir of hair follicle stem cells.

We speculate that dermal dendritic cells situated in the SMH might have a role in the hypertrichosis. However, the definite relation between SMH and hypertrichosis remains to be established.

Though it was focused one case to generalize the results, it is not possible to generalize the results. However, our case shows that a simple, non-invasive dermoscopic examination to determine hair density could be an ancillary method to predict the number of smooth muscle bundle in the dermis.

CONFLICTS OF INTEREST

The authors have nothing to disclose.

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A Case of Atrophoderma Vermiculatum Showing a Good Response to Topical Tretinoin

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Dear Editor:

Atrophoderma vermiculatum (AV) is a rare, slowly progressive, benign follicular disorder that affects primarily children¹. AV is characterized by the development of inflammatory, keratotic papules of the face that form pitted, atrophic, and depressed scars in a reticular or honeycomb pattern^{1,2}.

A 12-year-old girl presented with sudden onset of atrophic scarring of both cheeks. Examination showed multiple, pitted, honeycomb scars on the both cheeks, temples, chin and neck. The lesions were similar regarding their size and morphology, were oval shaped, skin-colored, 1 ~ 2 mm in diameter, and approximately 1 mm deep (Fig.

1A, B). The lesions had developed several years prior and had gradually expanded centrifugally. Neither the eyebrows nor eyelashes were involved; no scarring alopecia was evident. The patient denied any subjective symptoms including itching or pain. Her medical history contained atopic dermatitis but no evidence of any physical trauma or inflammation prior to disease onset. The physical examination and routine laboratory test showed all normal results. There was no relevant family history.

Histopathological examination of a punch biopsy specimen of an atrophic keratotic papule on the cheek revealed follicular hyperkeratosis; aberrant, atrophic pilosebaceous units formed small finger-like projections into the sur-

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