Acquired Dermal Melanocytosis Occurring in a Patient with Hypohidrotic Ectodermal Dysplasia

Dong Ju Hyun, Dong Hyun Kim, Moon Soo Yoon, Hee Jung Lee

Department of Dermatology, CHA Bundang Medical Center, CHA University, Seongnam, Korea

Dear Editor:

Hypohidrotic ectodermal dysplasia (HED) is characterized by a triad of clinical features including absent or diminished eccrine sweat glands, missing teeth, and sparse hair. Frequent skin manifestations of HED are xerosis and periorbital hyperpigmentation over the wrinkled skin, but there was no previous report of acquired dermal melanocytosis (ADM) in HED patient¹.

A 26-year-old man presented with bluish patches on the face, which developed one year ago. On physical examination, an alphabet X-shaped bluish patch on the glabella

and paranasal cheeks along Blaschko's lines and ovoid bluish patches on the temples were found (Fig. 1A). Typical clinical features of HED such as the characteristic face, dry skin, periorbital hyperpigmentation over the wrinkled skin, sparse hair and teeth were presented (Fig. 1). As he had sparse hair, he got a tattoo on the eyebrows. He also had a history of inability to sweat and recurrent febrile seizures since infancy and complained of high body temperature during the summer. He was diagnosed with atopic dermatitis in childhood. He had no family members showing similar manifestations.

Fig. 1. (A) Multiple bluish patches were observed on the glabella, both cheeks and both temples. Frontal bossing, depressed midface with a saddle nose, and everted lips were also noticed. The patient got a tattoo on the eyebrows for covering absent eyebrows. Periorbital hyperpigmentation over the wrinkled skin was also observed. (B) Hypotrichosis affecting axillary hair. (C) Oligodontia treated with dental prosthesis.

Received September 9, 2015, Revised October 28, 2015, Accepted for publication November 11, 2015

Corresponding author: Hee Jung Lee, Department of Dermatology, CHA Bundang Medical Center, CHA University, 59 Yatap-ro, Bundang-gu, Seongnam 13496, Korea. Tel: 82-31-780-5240, Fax: 82-31-780-5247, E-mail: derma97@cha.ac.kr

This is an Open Access article distributed under the terms of the Creative Commons Attribution Non-Commercial License (<http://creativecommons.org/licenses/by-nc/4.0>) which permits unrestricted non-commercial use, distribution, and reproduction in any medium, provided the original work is properly cited.

Copyright © The Korean Dermatological Association and The Korean Society for Investigative Dermatology

We performed a 3-mm-punch biopsy on the left paranasal cheek, which revealed flattened epidermis, hypoplasia of skin appendages and mild superficial perivascular lymphocytic infiltrations (Fig. 2A). In the dermis, multiple elongated, slightly wavy pigmented cells reacting with melan-A and S-100 were seen (Fig. 2B~D). The clinicopathological findings led to the diagnosis of ADM with HED. We suggested the patient to get genetic study but he refused because of the high cost. The pigmented lesions showed improvement after Q-switched neodymium-doped yttrium aluminium garnet (Nd:YAG) laser treatment. Periorbital hyperpigmentation in HED usually presents as grayish patch over fine wrinkles just like postinflammatory hyperpigmentation in atopic dermatitis². Because the pigmented lesion in our case presented as X-shaped bluish patch without wrinkles, it is clinically different from previously reported cases of periorbital hyperpigmentation in

HED. Although there are no biopsied cases of periorbital hyperpigmentation in HED, epidermal hypermelanosis and dermal melanophages may present in those cases like as histopathology of postinflammatory hyperpigmentation and it would be a differential diagnostic point.

The most frequent form of ADM is acquired bilateral nevus of Ota-like macules and other unusual forms of ADM were also reported³. Pathogenesis of ADM is poorly understood, but the faulty activation of dormant or inactive dermal melanocytes is considered as the main pathogenesis⁴. Various factors, such as ultraviolet (UV) rays, excessive sex hormone, trauma, chronic inflammation, interaction of collagen and elastic fibers or genetic factor can trigger ADM⁵. In our case, considering mild dermal inflammation seen in the pathology specimen, chronic inflammation is the most like triggering factor, but trauma from tattooing may also be involved. Furthermore, the ef-

Fig. 2. Biopsied specimen from the left cheek. (A) Flattened epidermis and hypoplasia of skin appendages was remarkable. Mild superficial perivascular lymphocytic infiltrations were noted (H&E, $\times 40$). (B) Multiple elongated, slightly wavy pigmented cells were seen in the dermis (H&E, $\times 100$; inset: H&E, $\times 400$). The pigmented cells reacted with melan-A (C) and S-100 (D) ($\times 200$).

fect of UV rays cannot be excluded.

As for most dermal pigmentary lesions, the Q-switched Nd:YAG laser is widely accepted treatment for the ADM. Our patient showed clinical improvement of the pigmentation after 8 sessions of Q-switched Nd:YAG laser treatment. Herein, we report a rare case of ADM on the glabella, paranasal cheeks and temples in a patient with HED.

REFERENCES

1. Itin PH, Fistarol SK. Ectodermal dysplasias. *Am J Med Genet C Semin Med Genet* 2004;131C:45-51.
2. Koguchi-Yoshioka H, Wataya-Kaneda M, Yutani M, Murota H, Nakano H, Sawamura D, et al. Atopic diathesis in hypohidrotic/anhidrotic ectodermal dysplasia. *Acta Derm Venereol* 2015;95:476-479.
3. Harrison-Balestra C, Gugic D, Vincek V. Clinically distinct form of acquired dermal melanocytosis with review of published work. *J Dermatol* 2007;34:178-182.
4. Hori Y, Takayama O. Circumscribed dermal melanoses. Classification and histologic features. *Dermatol Clin* 1988; 6:315-326.
5. Fauconneau A, Beylot-Barry M, Vergier B, Robert-Barraud C, Doutre MS. Acquired dermal melanocytosis of the back in a Caucasian woman. *Am J Dermatopathol* 2012;34:562-563.

<https://doi.org/10.5021/ad.2016.28.6.787>

Pseudoaneurysm as a Post-Biopsy Complication

Geo Han, Jung Woo Lee, Seung Hwi Kwon, Jiehyun Jeon, Hae Jun Song, Chil Hwan Oh, Sang Il Suh¹, Soo Hong Seo

Departments of Dermatology and ¹Radiology, Korea University Guro Hospital, Seoul, Korea

Dear Editor:

Dermatologists may encounter unexpected complications after skin biopsy. Cook and Perone¹ prospectively examined dermatologic surgical complications in the office setting, and overall complication rate of 1.64% in 1,343 cases were found, most of which was minor difficulties with hemostasis. Vascular complications rarely occur in dermatologic outpatient based procedures. It is thought to be especially rare after punch biopsies with relatively small and superficial incision. Complications of this category include

post-biopsy bleeding, slowly developing chronic expanding hematoma² and rarely pseudoaneurysm³. Here, we report a case of pseudoaneurysm after punch biopsy which

Received September 7, 2015, Revised October 29, 2015, Accepted for publication November 13, 2015

Corresponding author: Soo Hong Seo, Department of Dermatology, Korea University Guro Hospital, 148 Gurodong-ro, Guro-gu, Seoul 08308, Korea. Tel: 82-2-2626-1306, Fax: 82-2-2626-1309, E-mail: drsshong@hanmail.net

This is an Open Access article distributed under the terms of the Creative Commons Attribution Non-Commercial License (<http://creativecommons.org/licenses/by-nc/4.0>) which permits unrestricted non-commercial use, distribution, and reproduction in any medium, provided the original work is properly cited.

Copyright © The Korean Dermatological Association and The Korean Society for Investigative Dermatology

Fig. 1. A 1 × 1.1-cm-sized pulsating mass with intermittent pain.