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Acquired unilateral melanocytic nevi in otherwise normal skin

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Abstract

We describe a case with numerous melanocytic nevi in otherwise normal skin. A 5-year-old girl presented with more than 100 small pigment lesions on her left arm, shoulder and upper back without underlying light brown macule. The pigment lesions were first found on her left forearm at 3 months old, and gradually increased along with her growth. Skin biopsy from a pigmented lesion shows a pathological change of compound-type melanocytic nevus without any atypical changes. Speckled lentiginous nevus is known to have multiple melanocytic lesions on the underlying brown macule from birth. Partial unilateral lentiginosis is characterized by the unilateral lentigines with histopathological changes of lentigo or jentigo but not melanocytic proliferation in dermis. Agminated acquired melanocytic nevi usually occur in a narrow segmented area, but not in widely distributed area like this case. We consider that this is an unusual type of mosaicism of melanocytic disorders.

Introduction

Various types of unilateral multiple pigmented lesions in a Blaschko line area have been described. Speckled lentiginous nevus (SLN) is characterized by locally-disseminated multiple small pigment papules or macules which develop in the underlying well-demarcated light brown macule [1, 2]. The number of pigment lesions increases along with the growth, and the histology of them shows melanocytic nevi. Partial unilateral lentiginosis (PUL) is another form of disorder showing multiple pigmented lesions characterized by the unilateral segmental distribution of numerous lentigines in otherwise normal skin [3-5]. Its pathological feature is the typical lentigo, that is, melanocytic proliferation without nest formation in the basal layer of the epidermis. It occurs from birth time to the infant period. Here we report a female patient with segmentally-distributed numerous pigment lesions on otherwise normal skin, and one of the pigment lesions has histologically compound type melanocytic nevus.

Case report

A 5-year-old girl presented for the examination of numerous moles on her left arm, shoulder and back on Oct 14, 2005. The pigment lesions were not present from birth but found on the extensor surface of her left forearm when she was 3 months old. The number of the moles gradually increased and the extent of involvement covered her

left arm, left shoulder, and left upper back. She has neither developmental abnormality nor other disease. No other persons in her family had the similar symptom. On the examination of the first visit, there were numerous brown to dark brown macules or slightly elevated papules measuring 1 to 3 mm in diameter on her left forearm, upper arm, neck and upper back (Fig. 1A). The number of the pigmented lesions counted over 150 and the lesions were most frequently observed on extensor surface of her left forearm. There were pea-sized and nail-sized depigment macules, nevus achromicus, on her left axilla (Fig.1D). Otherwise, no other abnormality was found on her skin. At a following examination on January 5, 2006, a biopsy was taken from a dark brown papule on her left upper arm. The histology showed a typical compound nevus, showing nest formation of nevomelanocytes in the basal layer of the epidermis and in the upper dermis (Fig. 2). No atypia was observed. The number of pigmented lesions gradually increased and over 250 moles were observed in April 2007 (Fig. 1B and C).

Discussion

There are various types of multiple pigmented lesions that are localized to a certain area of the body; PUL, SLN, agminated acquired melanocytic nevi, and unilateral dysplastic nevi [1-6]. PUL is distinguished by small-circumscribed pigmented macules on otherwise normal skin in a unilateral part of the body. Upper part of the body including face, neck, shoulders, axillae, arms, chest and upper back, is frequently affected [5]. The pigmented lesions appear in 1 to 15 years after birth. A histopathological change of PUL is a typical lentigo simplex that shows an increase of nevoid melanocytes in the basal layer of the epidermis. In a rare occasion, PUL has been reported to show a histological finding of “jentigo” which show a small cluster of nevomelanocytic nest in the basal layer [8]. In the present case, histopathology of the biopsy specimen showed the typical compound-type nevus. We therefore consider that this case is distinct from PUL.

SLN shows a number of pigmented flat or elevated lesions scattered on a widely distributed brown macule, which usually exists at birth [1]. Recently, two types of SLN have been proposed, that is, macular and papular types [2]. Macular SLN is characterized by a tannish-brown background with darker flat speckles that histologically present jentigo pattern like PUL, while papular SLN shows a light-brown macule with multiple small melanocytic nevus. The small pigmented lesions in the

underlying brown macule may exisit at birth and frequently increase after birth. They usually range from 1 to 3 mm in diameter. Concerning histological picture of scattered pigmented lesions, the present case resembles papular SLN, however we could not observe any light-brown macules at the area where the multiple pigment lesions exist. We therefore consider that the present case is not compatible with SLN.

Agminated acquired melanocytic nevi (AAMN) may occur in a segmented area [6, 9]. However, AAMN is usually observed within a narrow segmented area, but not in widely distributed area, such as whole arm and back as in this case. Banuls et al reported a 15-year-old girl who had grouped congenital melanocytic nevi on her left preauricular region and acquired linearly-arranged melanocytic nevi on her left nuchal area [10]. The pigmented lesions were 0.6-2 cm in diameter. Other reported cases with congenital unilateral multiple melanocytic nevi had pigmented lesions that measured 0.1 to 3 cm in diameter [11, 12]. These all cases showed pigmented lesions at birth. The present case has much smaller melanocytic nevi that measured less than 3 mm in diameter and developed after 3 months old. Thus, this case exhibits a unique unilateral multiple melanocytic nevi that appeared after birth. Unilateral dysplastic nevi associated malignant melanoma has been described in a Japanese [7]. The moles measuring 4 to 5 mm in diameter developed unilaterally after birth in otherwise normal skin. The present case may mimic unilateral dysplastic nevi, however lack of irregular borders in moles and absence of atypia are not consistent with this disorder.

The clinical feature of unilaterally scattered small pigmented lesions on an otherwise normal skin is similar to PUL, and the histological feature seems to be close to SLN. Thus, the present case has the feature of these two pigment disorders. It has been proposed that SLN may be associated with neurological abnormalities including sensory neuropathy, motor neuropathy, nerve palsy, muscular atrophy and hyperhidrosis [13, 14]. The present case has no neurological abnormality but should be carefully observed further. It has been suggested that SLN has an increased risk of developing malignant melanoma [15-17]. Higher number of acquired melanocytic nevi has been shown to be a risk factor for melanoma [18]. Further, it has been shown that malignant melanoma developed in a patient with unilateral dysplastic nevi [7]. Therefore, the increased number of melanocytic nevi in a unilateral part of the body in this case suggests a higher risk of developing malignant melanoma.

Recently, the responsible genes have been identified in genetic disorders with

multiple lentigines such as Carney complex and LEOPARD syndrome [19]. These genes are related to the cellular proliferation. It appears that mosaic skin diseases develop by the loss of heterozygosity [20]. It is thus possible that disorders with unilateral multiple pigmented lesions including PUL, SNL and this case are associated with loss of heterozygosity of the appropriate genes in the segemental part of the body.

References

1. Schaffer JV, Orlow SJ, Lazova R, Bolognia JL: Speckled lentiginous nevus: within the spectrum of congenital melanocytic nevi. Arch Dermatol 2001;137:172-8.
2. Vidaurri-de la Cruz H, Happle R: Two distinct types of speckled lentiginous nevi characterized by macular versus popular speckles. Dermatology, 2006;212:53-58.
3. Kang S, Sober AJ: Disturbances of melanin pigmentation; In Moschella SL, Hurley HJ (eds): Dermatology. Philadelphia. WB Saunders, 1992, pp 1442-5.
4. Romiti R, Harnache JDI , Neto CF, Kreuter A, Altmeyer P, Jansen T: Extensive partial unilateral lentiginosis. J Dermatol 2001;28:490-2.
5. Trattner A, Metzker A: Partial unilateral lentiginosis. J Am Acad Dermatol 1993;29:693-5.
6. Bragg JW, Swindle L, Halpern AC, Marghoob AA: Agminated acquired melanocytic nevi of the common and dysplastic type. J Am Acad Dermatol 2005;52:67-73.
7. Misago N, Takahashi M, Kohda H: Unilateral dysplastic nevi associated with malignant melanoma. J Dermatol 1991;18:649-53.
8. Marchesi L, Naldi L, Di Landro A, Cavalieri d'Oro L, Brevi A, Cainelli T: Segmental lentiginosis with "jentigo" histologic pattern. Am J Dermatopathol 1992;14:323-7.
9. Marghoob AA, Blum R, Nossa R, Busam KJ, Sachs D, Halpern A: Agminated atypical (dysplastic) nevi: case report and review of the literature. Arch Dermatol 2001;137:917-20.
10. Bañuls J, Ramon R, Guijarro J, Alfonso R, Silvestre JF, Climent JM, Botella R: Segmental arrangement of multiple, partly congenital and partly acquired melanocytic nevi. Eur J Dermatol 1998;8:80-2.
11. Effendy I, Happle R: Linear arrangement of multiple congenital melanocytic nevi. J Am Acad Dermatol 1992;27:853-4.
12. Lin YT, Teng RJ, Chiu HC, Yau KI, Chang MH, Shan YZ: An unusual presentation of multiple congenital melanocytic nevi with a limb distribution. Dermatology 1997;194:362-3.
13. Happle R: Speckled lentiginous nevus syndrome: delineation of a new distinct neurocutaneous phenotype. Eur J Dermatol 2002;12:133-5.
14. Vente C, Neumann C, Bertsch H, Rupprecht R, Happle R: Speckled lentiginous

nevus syndrome: report of a further case. Dermatology 2004;209:228-9.

15. Bolognia JL: Fatal melanoma arising in a zosteriform speckled lentiginous nevus. Arch Dermatol 1991;127:1240-1.
16. Kurban RS, Preffer FI, Sober AJ, Mihm MC, Barnhill RL: Occurrence of melanoma in "dysplastic" nevus spilus: report of case and analysis by flow cytometry. J Cutan Pathol 1992;19:423-8.
17. Rhodes AR, Mihm MC: Origin of cutaneous melanoma in a congenital dysplastic nevus spilus. Arch Dermatol 1990;126:500-5.
18. Rokuhara S, Saida T, Oguchi M, Matsumoto K, Murase S, Oguchi S: Number of acquired melanocytic nevi in patients with melanoma and control subjects in Japan: Nevus count is a significant risk factor for nonacral melanoma but not for acral melanoma. J Am Acad Dermatol 2004;50:695-700.
19. Bauer AJ, Stratakis CA: The lentiginoses: cutaneous markers of systemic disease and a window to new aspects of tumourigenesis. J Med Genet 2005;42:801-10.
20. Frank J, Happle R: Cutaneous mosaicism: right before our eyes. J Clin Invest 2007;117:1216-9.

Figure legends

Fig. 1. Clinical appearance of unilateral melanocytic nevi in otherwise normal skin on left upper back, shoulder and arm (A). The number of pigment lesions gradually increased, and over 250 moles were observed on April 5, 2007 (B and C). Small pigment lesions on the left upper arm. Pigment lesions measured 1 to 3 mm in diameter. Neither irregular borders nor color variegation in moles were observed.

Fig. 2. Histology of a black papule on left upper arm, showed a typical compound nevus. Nest formation of nevomelanocytes in both basal layer of the epidermis and the upper dermis was shown. No atypia was observed (hematoxylin-eosin stain). The bar indicates 50μm.



Fig. 1

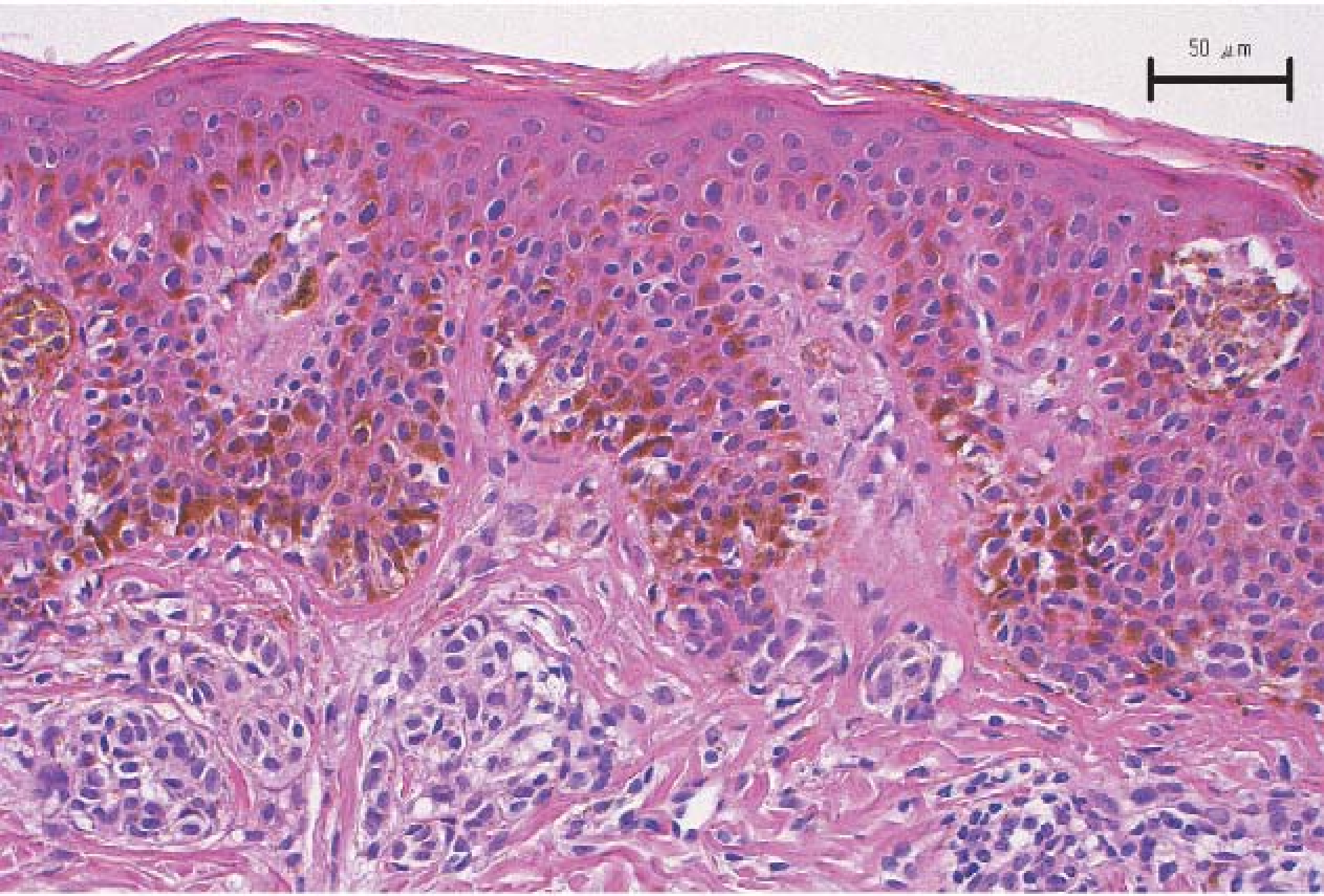


Fig. 2