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Eruptive facial lentiginosis-like repigmentation in a patient with longstanding generalized vitiligo without a detectable trigger

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Abstract

Spontaneous appearance of hyperpigmented macules on chronic vitiligo lesions is a very rare phenomenon, which is described as eruptive lentiginosis. We describe the case of a patient with chronic non-segmental generalized vitiligo who presented with a sudden onset of hyperpigmented macules on depigmented areas of the face. A biopsy showed pigmented basal keratinocytes in the interfollicular epidermis, and immunohistochemistry with anti-SOX10 antibodies showed nuclei of single melanocytes. This case shows that even long-standing depigmented vitiligo lesions may contain functional melanocytes or their precursors.

Keywords: vitiligo, generalized, repigmentation, eruptive, lentiginosis-like pattern, SOX10-positive melanocytes

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Introduction

Vitiligo is an autoimmune disease manifesting as skin depigmentation due to a loss of functional melanocytes (1). Spontaneous repigmentation in the form of eruptive lentigines is very rarely described in patients with longstanding vitiligo (2, 3).

Case report

We report the case of a 56-year-old man with non-segmental generalized vitiligo that presented due to the sudden onset of facial hyperpigmented macules. The initial pigmented spots appeared 2 months prior to the presentation on the skin beneath his eyes and then spread to the nose, forehead, ears, and cheeks. The patient has been suffering from vitiligo since early childhood with ongoing slow depigmentation over the years. The last treatment attempt spanned 5 years and comprised 300 sessions of narrowband ultraviolet (UV) B phototherapy (three times weekly) and topical tacrolimus. Only limited and short-lived repigmentation was achieved. He is otherwise healthy, taking no systemic medications. He did not report increased sun exposure; the average UV index in the patient's area of residence around the time the lesions appeared is 3. The lesions were dark brown macules coalescing into patches on his cheeks, forehead, nose, temples, and ears (Fig. 1A, B). Dermoscopically, on a homogenous light to dark brown background, lighter follicular openings were seen (Fig. 1C). Vitiligo was generalized, affecting the face, as well as large areas on the trunk, legs (Fig. 1D), and arms. A biopsy was taken from the

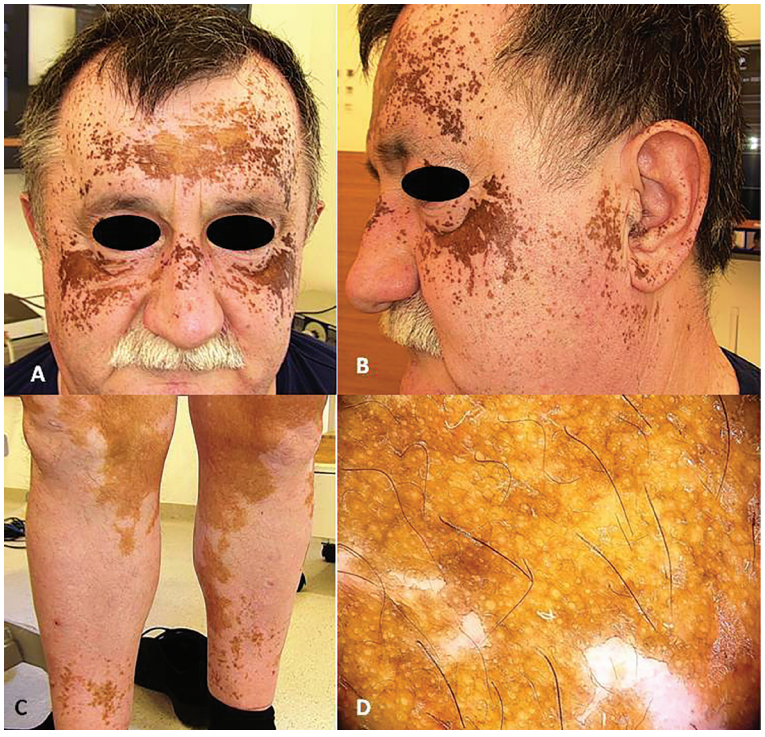

Figure 1 | Tanned coalescing macules on the (A) frontal and (B) left side of the face; (C) vitiliginous depigmentation affected almost the entire lower legs; (D) dermoscopy of the lentiginous repigmentation in the patient: a diffuse light brown background with a negative network and darker brown contouring of clear hair follicles.

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cheek and stained with hematoxylin and eosin (H&E) along with immunohistochemistry with the anti-SOX10 antibody in a search for epidermal melanocytes (Fig. 2A–C). H&E sections revealed heavily pigmented basal keratinocytes of the interfollicular epidermis (Fig. 2A and B) and anti-SOX10-stained nuclei of single melanocytes (Fig. 2C).

Discussion

Eruptive lentiginosis, either circumscribed or generalized, may be part of various syndromes or an acquired condition, usually occurring during chemotherapy or other immune-modifying treatments (4, 5). It is thought that weakening of the immune system allows melanocytes to proliferate and form lentigo macules. In rare cases, no obvious trigger may be discerned, as in cases of generalized eruptive lentiginosis (6). Depending on the methods of detection, studies found conflicting results regarding the survival of melanocytes in vitiliginous lesions (3, 7). In our practice, SOX10 staining was consistently negative in fully depigmented vitiligo lesions (not shown). However, the appearance of lentigines in vitiligo, at least in some patients, supports the hypothesis that there are residual, dormant melanocytes even in longstanding depigmented areas. Usually, the sudden development of lentigo macules in vitiligo patients follows some form of inflammatory insult or immune-modifying agent, such as psoralen and UVA photochemotherapy, intense sun exposure, or azathioprine administration (2, 5, 8). We could not identify any of these factors in our patient. Repigmentation in vitiligo can start from hair follicles, from unaffected borders, from the interfollicular epidermis, or as a combined pattern (9, 10). We could not find dermoscopic images of published cases of lentigines arising in vitiligo lesions, but in our patient it is clear that SOX10-positive melanocytes are mostly distributed in the interfollicular epidermis—hence the negative pigment network with hypopigmented holes representing hair follicles, which is a typical dermoscopic feature of solar lentigines. This might correspond to the diffuse pattern of repigmentation. However, the pigmented macules in the patient cannot be labeled "lentigines" because there is no increase in the number of melanocytes but only an increase in melanin production and deposition within keratinocytes. Other features of solar lentigo are missing, such as club-shaped rete ridges and solar elastosis. The lack of melanophages in the dermis, necrotic keratinocytes, or lymphocyte exocytosis excludes a significant inflammatory component as seen in previously reported cases of "lentiginous" repigmentation in vitiligo (5). The patient shows that even fully depigmented vitiligo lesions over 30 years old may contain functional melanocytes (or their precursors) in the interfollicular epidermis. They may be activated and produce large amounts of melanin.

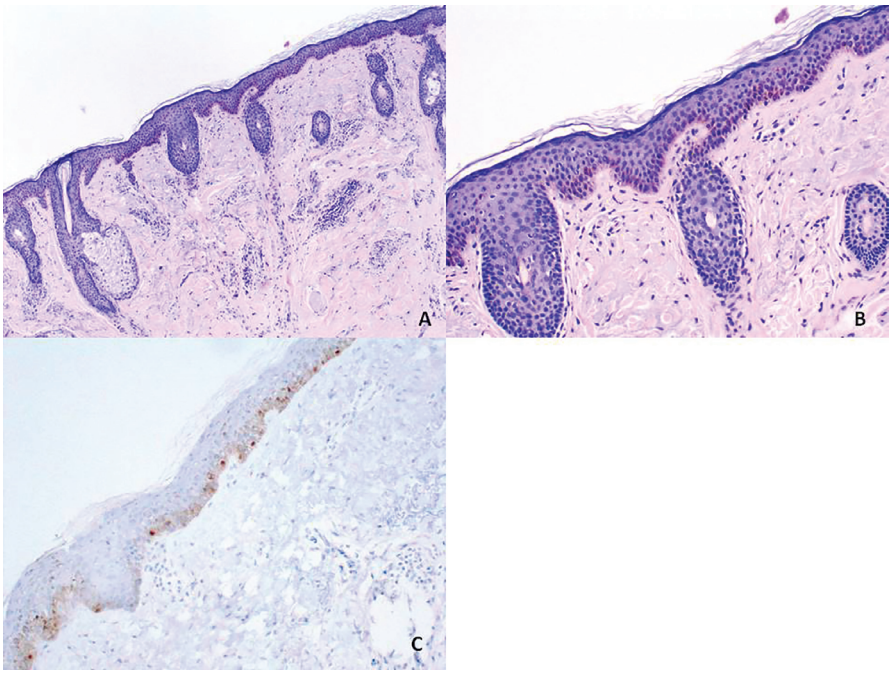

Figure 2 | (A, B) Heavy pigmentation of keratinocytes of the interfollicular epidermis (H&E, 10× and 20×); (C) SOX10-positive nuclei of melanocytes seen as single cells along the basement membrane of the interfollicular epidermis (20×, counterstained with Mayer's hematoxylin).

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