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BRIEF REPORT

Unilateral Linear Keratosis Pilaris on Hypopigmented Patches: An Additional Case of a New Variant of Keratosis Pilaris in an Asian Male

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Dear Editor:

Keratosis pilaris (KP) is a common inherited disorder characterized by small, folliculocentric keratotic papules with surrounding erythema[1]. KP typically occurs bilaterally and preferentially on the extensor surfaces of the upper arms, thighs, and buttocks[2]. In this study, we report a case of KP displaying a unilateral linear distribution, potentially associated with cutaneous mosaicism.

A 13-year-old boy presented with hypopigmented patches containing linear follicular papules on the right flank. Hypopigmented patches had appeared on his right flank one year prior (Fig. 1A). After several months, linear follicular papules developed on these hypopigmented patches (Fig. 1B, C). The patient's father also had typical KP on the bilateral extensor surfaces of the upper arms, buttocks, and upper thighs. The past medical history of our patient was unremarkable. Histopathological analysis of a follicular papule revealed hyperkeratosis and a dilated follicular infundibulum with keratotic plugging (Fig. 2A). Moreover, the analysis of two biopsies taken from a hypopigmented patch and normal perilesional skin demonstrated decreased melanin content in the hypopigmented patch (Fig. 2B, C).

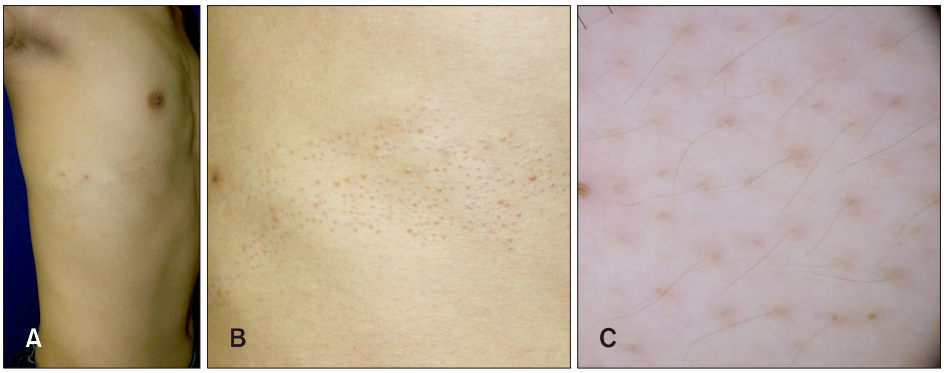

Fig. 1. (A, B) Linear follicular papules on hypopigmented patches of the right chest wall. (C) Dermoscopic visualisation of follicular papules.

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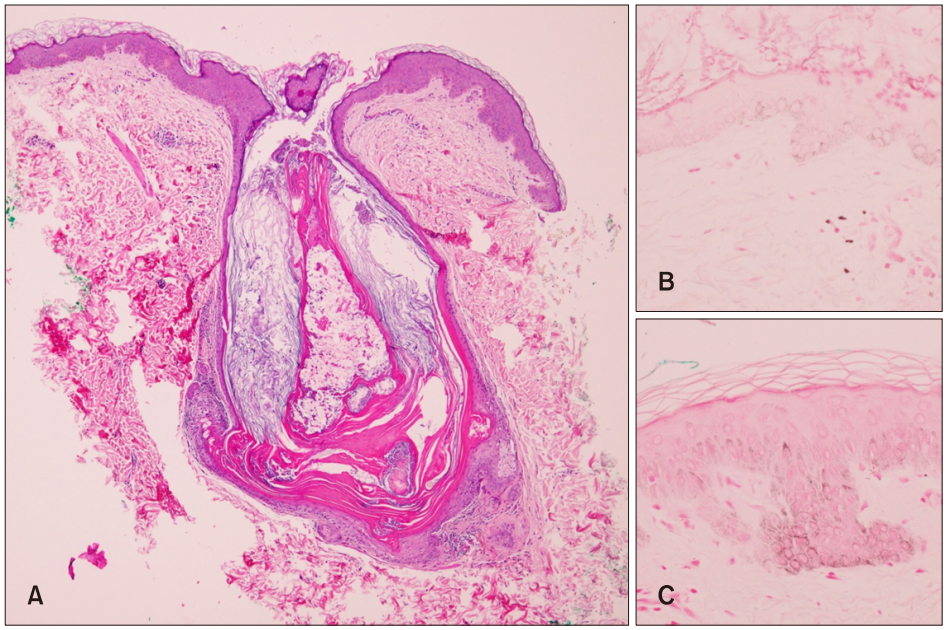

Fig. 2. (A) Hyperkeratosis and dilated follicular infundibulum with keratotic plugging (H&E, ×40). Reduced melanin content in hypopigmented patch (B) compared to normal skin (C) (Fontana–Masson stain, ×200).

Based on these clinico-histopathological features, we made the diagnosis of unilateral linear KP on hypopigmented patches.

There have been several reports of unilateral KP[2-4]. Ehsani et al.[2] and Zhu et al.[3] reported cases of generalized unilateral KP. These authors suggested that the unilateral distribution of their cases could be explained by hormonal change or genetic mutations[2,3]. Similar to the present case, Ma et al.[4] reported a case of localized unilateral KP occurring on pre-existent hypopigmented patches of the right flank. The authors suggested that a mosaic mutation of the fibroblast growth factor receptor 2 (*FGFR2*) gene may be associated with the observed pathogenesis[4,5]. According to the hypothesis of Ma et al.[4], *FGFR2* gene mutation in keratinocytes is sufficient to induce hypercornification of the pilosebaceous duct and an inflammatory response. Furthermore, the presence of hypopigmentation may be associated with the role of *FGFR2* signalling in skin pigmentation processes, specifically the melanocyte–keratinocyte interaction[4,5]. Although, the exact pathogenic mechanisms of KP have not been fully elucidated, Filaggrin (*FLG*) gene mutation has been proposed for the pathogenesis of KP[1,4]. However, *FLG* gene mutation could not explain skin hypopigmentation in our case. Therefore, as Ma et al.[4] proposed, it needs to be considered that postzygotic mutation in the *FGFR2* gene might be potentially play a role in the development of lesions in our patient.

In this study, we presented an additional case of a new mosaic variant of KP displaying a linear distribution on hypopigmented patches[4]. Further genetic studies are needed to determine the exact pathogenesis associated with the peculiar clinical features of this disorder. We received the patient's consent form about publishing all photographic materials.

CONFLICTS OF INTEREST

The authors have nothing to disclose.

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