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Case Study

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A RARE CASE: ONE-SIDED RETICULATE ACRAL PIGMENTATION OF KITAMURA

[1]Dr. Rajkiran Takharya, [2]Dr. Jude E. Dileep, [3*]Dr. P. Oudeacoumar, [4]Dr. Ilakkia P. Sadasivam, [5]Dr. Lisa J. D'souza, [6]Dr. Bhavana Sampadarao, [7]Dr. Divya Mani and [8]Dr. Gayathri Jayabalan

[1,4,5]3rd Year Postgraduate, Department of Dermatology, Aarupadai Veedu Medical College and Hospital, Puducherry, India.
[2]Assistant Professor, Department of Dermatology, Aarupadai Veedu Medical College and Hospital, Puducherry, India.
[3]Professor and Hod, Department of Dermatology, Aarupadai Veedu Medical College and Hospital, Puducherry, India.
[6,7,8]2nd Year Postgraduate, Department of Dermatology, Aarupadai Veedu Medical College and Hospital, Puducherry, India.

***Corresponding Author: Dr. P. Oudeacoumar**

Professor and Hod, Department of Dermatology, Aarupadai Veedu Medical College and Hospital, Puducherry, India.

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ABSTRACT

Reticulate acropigmentation of Kitamura (RAPK) is a less common genodermatosis characterized by pigmented, angulated, atrophic freckle-like lesions arranged in a reticulate pattern over the dorsal surface of the hand. We herein report a case of unilateral RAPK in an 18-year-old girl. The patient has cutaneous lesions and histopathology characteristic of RAPK, involving only the right hand. Because of its rarity, this case has been registered.

KEYWORDS: Acropigmentation, Genodermatosis, Reticulate Pigmentation, Unilateral.

INTRODUCTION

Reticulate pigmentary disorders are rare genetic abnormalities generally inherited in an autosomal dominant manner. These disorders include reticulate acropigmentation of Dohi (RAPD), reticulate acropigmentation of Kitamura (RAPK), Haber's syndrome, Dowling–Degos disease (DDD), and Galli–Galli disease.[1]

RAPK is characterized clinically by reticulate, slightly depressed pigmented macules on the acral parts of the body (mainly the dorsum of the feet and hands). Pits and breaks are located at the palms, soles, and dorsal phalangeal surfaces.[2] The lesions begin in the first and second decades of life and steadily increase onto the extremities and seldom on the face and eyelids. The lesions typically darken slowly over time.[3]

CASE REPORT

An 18 -year-old lady came to OPD with asymptomatic, darkish coloured skin lesions on the dorsum of the right hand for two months with no other complaints. The lesions were first less in number then progressed proximally to involve most of the right hand and forearm. The colour of lesions darkened with time from light brown to darkish brown. There was no history of chronic intake of any drugs, trauma or photosensitivity.

Dermatological examination revealed hyperpigmented, angulated, atrophic macules in a reticulate pattern involving only the right side of her hand and forearm (Figure 1). The ventral surface of the same hand was not involved (figure 2). Pits with breaks in dermatoglyphics were absent on both the palms. Other body parts like soles, scalp, hair, nails, teeth, Mucous membranes were normal. Cutaneous biopsy of a hyperpigmented macule over the right dorsum of the hand revealed thinning of the epidermis, elongation of rete ridges with increased number of melanocytes in the basilar keratinocytes (Figure 3). A diagnosis of unilateral reticulate acral pigmentation of Kitamura was made.

Table 1: Differential diagnosis of reticulate acropigmentation of Kitamura Salient features.

Disorders	Salient and associated features
RAPD (Reticulate acropigmentation of Dohi)	Mottled pigmentation with patchy hypopigmentation or depigmentation present over mainly over dorsal extremities. Freckle like macules may present over facial area.
Dyskeratosis congenital	Reticulate hyperpigmentation with poikiloderma of the thighs, chest, and neck. Other features include leukoplakia oral mucous membrane and dystrophy of nail.

Naegeli–Franceschetti–Jadassohn syndrome	Reticulate pigmentation often involving axillae and neck. Keratoderma of the soles and palms is usual. Associated with hypohidrosis.
Dyschromatosis universalis hereditaria	Symmetrical distribution of hypopigmented and hyerpigmented macules on the whole body. Associated with small stature and high tone deafness.
Dermatopathia pigmentosa reticularis	Prominent Reticulate hyperpigmentation over the trunk with mucosal involvement. Associated with Onychodystrophy and alopecia
DDD (Dowling–Degos disease)	Reticular hyperpigmentation over flexural areas with perioral pits and epidermal cyst. histology shows pigmented filiform epidermal projection.

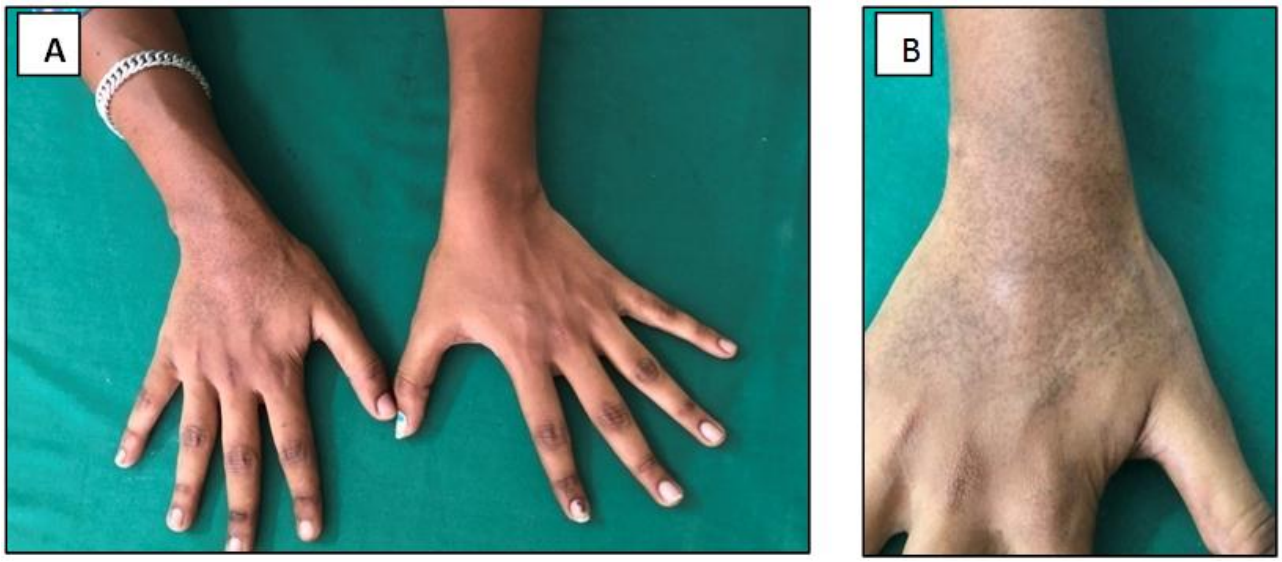

Figure 1: (a) Patchy reticulate angulated pigmented macules on the dorsum of the right hand. (b) Magnified image of dorsum of right hand.

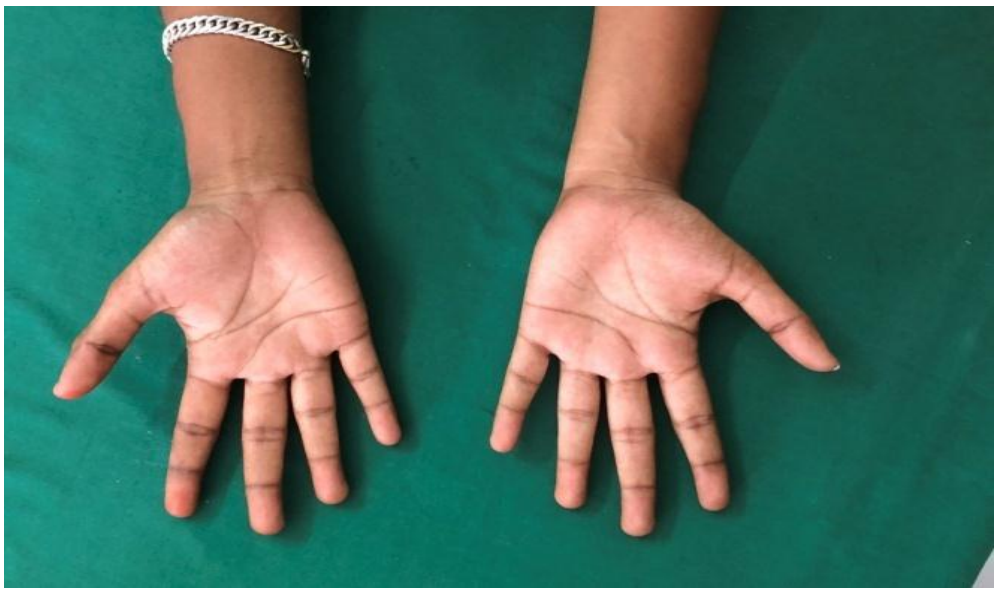

Figure 2: Ventral aspect of both palms without any pitting.

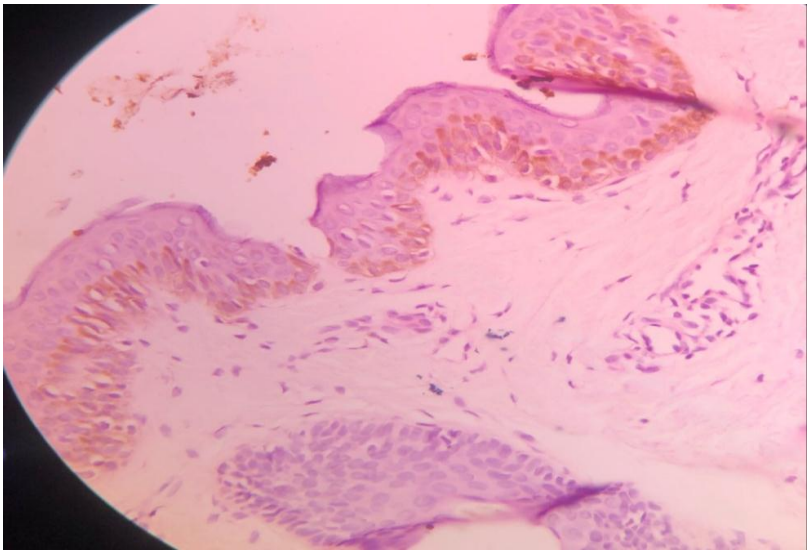

Figure 3: Thinning of epidermis, elongation of rete ridges with increased number of melanocytes in the basilar keratinocytes (H&E 40X).

DISCUSSION

Kitamura and Akamatsu from Japan originally described RAPK.[4] Close to one hundred cases are reported, principally in dark-skinned people of Asian ethnic origin.[5] RAPK is clinically characterized by hyperpigmented, angulated, and slightly atrophic macules over the distal extremities.[2] On histopathology will reveal atrophic epidermis with club-like elongation of the rete ridges and an excess of melanin in the basal layer.[5]

The differential diagnosis of RAPK includes RAPD, dyskeratosis congenita, Naegeli–Franceschetti–Jadassohn syndrome, dyschromatosis universalis hereditaria, dermatopathia pigmentosa reticularis and DDD (Table 1).[1] The differentiation between RAPD and RAPK clinically is in RAPD, there are hypopigmented macules. Hyperpigmentation on flexor areas are seen in DDD, reticular, and under histopathological examination, pigmented filiform projections in the epidermis with involvement of the follicular infundibulum are seen.[6]

Some authors have proposed that RAPK and DDD may be a continuous spectrum of a single entity, as various cases with overlapping features have been reported.[7,8] However, molecular research show mutations of KRT5 and POGLUT1, particularly in patients of DDD[9], whereas patients of RAPK are unveiled to have mutations in ADAM10.[10] Still, some of the patients with overlapping features of RAPK and DDD had mutations in the POFUT1 gene.[11]

Our patient had cutaneous lesions, which are classically observed in RAPK, and the skin biopsy confirmed the diagnosis of RAPK. Notably, our patient had a unilateral distribution of the lesions, which is exceptional in RAPK. This made us come to conclude that condition makes our patient become is a case with unilateral acral reticulate pigmentation of Kitamura. Treatment of this condition is often found to be unsatisfactory. Treatments that have been tried include 20% azelaic acid[3] and Q-switched alexandrite laser(755 nm).[12]

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