Idiopathic Eruptive Macular Pigmentation in an Indian Male

Sir,

We report a 24-year-old man who presented with asymptomatic dark brown lesions over the face and trunk of 20 years duration. They appeared spontaneously without any preceding erythema or topical/systemic therapy. The lesions started insidiously and gradually progressed over a period of 3 years followed by a quiescent phase of around 16 years and aggravation 1 year before presenting to us. The general physical and systemic examination was unremarkable. Cutaneous examination revealed multiple brownish-gray to dark, discrete, round-to-oval, barely elevated plaques of size 0.5 to 2 cm [Figure 1a and b]. Most of the lesions had a velvety texture. Palms and soles were spared. He also had mild acanthosis nigricans of bilateral axillae [Figure 1c].The mucosae, hair, and nails were normal. Darier's sign was negative. Hematological and biochemical investigations including fasting blood sugar. glycosylated hemoglobin, and fasting insulin levels were normal. Skin biopsy showed irregular acanthosis, papillomatosis [Figure 2], keratin horn formation, and

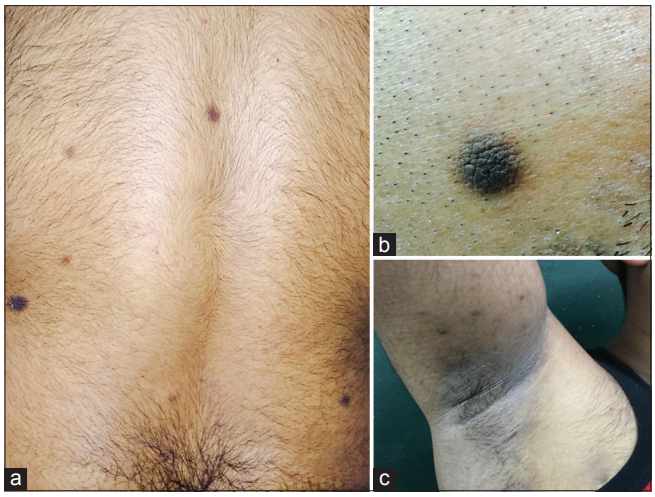

Figure 1: (a) Multiple hyperpigmented plaques on the trunk. (b) Close up picture showing velvety hyper pigmented plaque. (c) Acanthosis nigricans in axilla

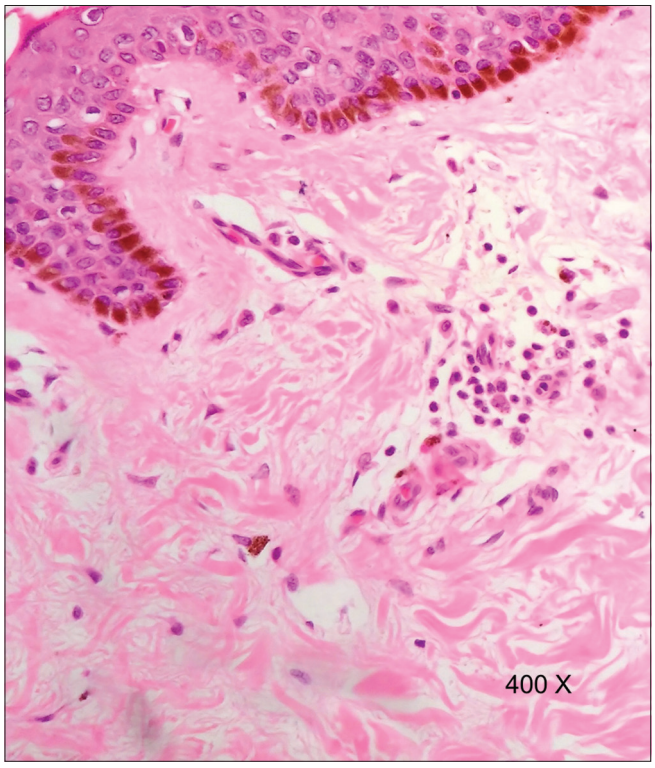

Figure 3: Skin biopsy H&E stain showing prominent basal layer hyper pigmentation

basal layer hyperpigmentation [Figure 3]. The upper dermis showed sparse superficial lymphohistiocytic infiltrate. Few melanophages were seen in the papillary dermis. The mast cell number was normal on hematoxylin and eosin stain. A diagnosis of idiopathic eruptive macular pigmentation (IEMP) was made based on characteristic clinical and histopathological features.

The patient was started on topical tretinoin 0.05% application once daily for 2 months without much improvement, following which it was discontinued.

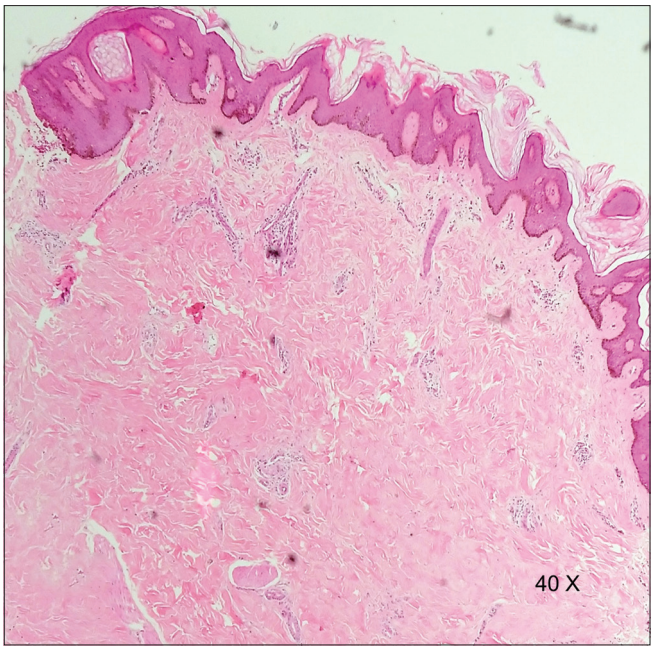

Figure 2: Skin biopsy H&E stain showing irregular acanthosis, papillomatosis, keratin horn formation, and basal layer hyperpigmentation

IEMP is a rare disorder of pigmentation of unknown etiology. It was first described by Degos *et al*.[1] in 1978, and since then, less than 50 cases have been reported. Joshi *et al*.[2] have reported the largest series of 48 cases. IEMP is self-resolving and has been reported to disappear spontaneously in months to years. An unusual case IEMP lasting for 21 years in a 24-year-old woman was characterized by several periods of spontaneous resolution followed by recurrences.[3]

The diagnostic criteria for IEMP were given by Sanz de Galdeano *et al*. in 1996 which includes (a) Eruption of brownish, nonconfluent, asymptomatic macules involving the trunk, neck, and proximal extremities in children and adolescents; (b) absence of preceding inflammatory lesions; (c) no previous drug exposure; (d) basal layer hyperpigmentation of the epidermis and prominent dermal melanophages without visible basal layer damage or lichenoid inflammatory infiltrate; (e) normal mast cell count (4).[4] Our patient fulfilled all these criteria.

Recently, lesions occurring in a Christmas tree pattern[5] and limited to flexures[6] have been described. The differential diagnosis include lichen planus pigmentosus (LPP), erythema dyschromicum perstans (EDP), fixed drug eruption, and mastocytosis. However, none of these have velvety lesions. In IEMP, a preceding cause is absent and Darier's sign is negative. The disease occurs primarily during childhood and adolescence usually without a history of erythema or drug medication unlike FDE. LPP is characterized by hyperpigmented, dark-brown macules in sun-exposed areas and flexural

Table 1: Cases published till date with histological emphasis on pigmented papillomatosis

Number of patients	Age of onset (years)	Site	Size	Duration (years)	Basal cell melanization	Pigmented Papillomatosis	Year (Reference)
5	2-16	Trunk, neck, arms, legs	3-25 mm	1-4	+	+ (mixed for different cases)	1996 (4)
1	24	Trunk, arms		21	+	-	2003 (3)
1	31	Trunk, thigh, forearm	2-7 cm	2	+	-	2005 (8)
9	6-14	Face, trunk, arms, legs	Up to 3 cm	2	-	+	2007 (9)
1	23	Axilla, cubital fossa, inguinal area		3	+	-	2008 (6)
1	21	Face, trunk, arms, legs	0.5-2 cm	2	+	+	2010 (7)
2	7-13	Face, trunk, arms, legs	5-15 mm	4-8 months	+	+	2010 (10)
1	10	Trunk, arms, legs	1-3 cm	3 months	+	+	2011 (11)
1	22	Trunk		1	+	-	2013 (5)
1	11	Face, trunk, arms, legs	0.5-3 cm	3 months	+	+	2014 (12)
1	6	Distal extremities			+	+	2014 (13)
1	8	Face, trunk, arms, legs	2-15 mm	3 months	+	+	2015 (14)
1	5	Neck, trunk, legs		10 months	+	+	2016 (15)
1	7	Face, trunk, arms, legs		7	+	+	2016 (16)
1	19	Face, trunk		6 months	+	-	2016 (17)
1	20	Face, trunk, arms, legs	0.5-2 cm	1.5	+	+	2016 (18)

folds whereas EDP has hyperpimented macules surrounded by erythema.

Histologically, an atrophic epidermis, a vacuolar alteration of the basal cell layer with a scarce lymphohistiocytic lichenoid infiltrate and pigment incontinence are seen in LPP. In EDP, slate gray macules with rim of erythema without any predilection for photoexposed sites are seen and pigment is also present in deep dermis. However, in IEMP, there is absence of lichenoid infiltrate or basal cell damage and mast cells are normal in number. There is acanthosis, basal layer hyperpigmentation, and dermal melanophages. The histopathological finding "pigmented papillomatosis," which is a characteristic feature reported in a series of nine cases from India was present in our case too.[7] Emphasis has been laid on the histological presence of pigmented papillomatosis to be a diagnostic criteria for IEMP [Table 1].[4-18]

This case is reported for its rarity with an objective to increase its awareness among dermatologists and pathologists. Although it has been called as eruptive acanthosis nigricans in view of clinical and histological similarity, there was no metabolic derangement in our patient. He had associated acanthosis nigricans of axillae which may suggest a possible relationship between these disorders. The clinical course of our patient was different as he never had spontaneous resolution but his disease stopped progressing followed by a sudden aggravation 16 years later.

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Conflicts of interest

There are no conflicts of interest.

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