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Case Series

Clinical and Histopathological Insights into Adult-Onset Unilateral Nevus of Ota

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ABSTRACT

Dermal melanocytoses are characterised by the presence of spindle-shaped melanocytes in the dermis. The common forms, such as Mongolian spots, and nevi of Ota or Ito, present from birth or puberty, while acquired dermal melanocytoses appear in adulthood and are rare. They are frequently misdiagnosed as lichen planus pigmentosus or melasma leading to delay in treatment. This study reports five cases of acquired unilateral nevus of Ota from India, with an analysis of their dermoscopic and histopathological features.

Keywords: Acquired bilateral nevus of Ota-like macules, Dermal melanocytosis, Dermoscopy, Hori's nevus, Nevus of Ota

INTRODUCTION

Dermal melanocytoses are a group of diverse entities with the common theme of an intradermal population of dendritic spindle shaped melanocytes. The presentation can be since birth or during puberty for Mongolian spot, Nevus of Ota (NOO) or Nevus of Ito. While Mongolian spot disappears soon after birth, NOO and Ito tend to persist. A second categorisation is the Acquired Dermal Melanocytosis (ADM) which presents during adulthood and is extremely rare. These include Hori's nevus or acquired bilateral nevus of ota like macules (ABNOM) and acquired NOO or the Sun's nevus. We present a series of five patients with histopathologically proven acquired unilateral NOO, along with their dermoscopic characteristics.

CASE SERIES

Four Indian females and one male with a mean age of 42 years presented with unilateral distribution of bluish-brown discrete to coalescing macules on an otherwise unremarkable face [Figure 1a-f]. The mean duration of the condition was 6 years. There was no history of appearance of lesions during pregnancy, drug intake, preceding trauma or topical application of cosmetic agents before onset of lesions. Other than bluish scleral pigmentation on the same side [Figure 1d] in all five patients, rest ophthalmological examination was normal. Dermoscopic examination was performed using a polarised handheld dermoscope with ×10 magnification (DermLite DL5, San Juan, Capistrano, CA, USA) [Figure 2a]. Histopathological examination was done in all cases and consistently showed presence of dermal melanocytes with long dendritic processes and cytoplasmic melanin pigment between collagen present in the mid and/or deep dermis [Figure 2b and c]. The clinical,

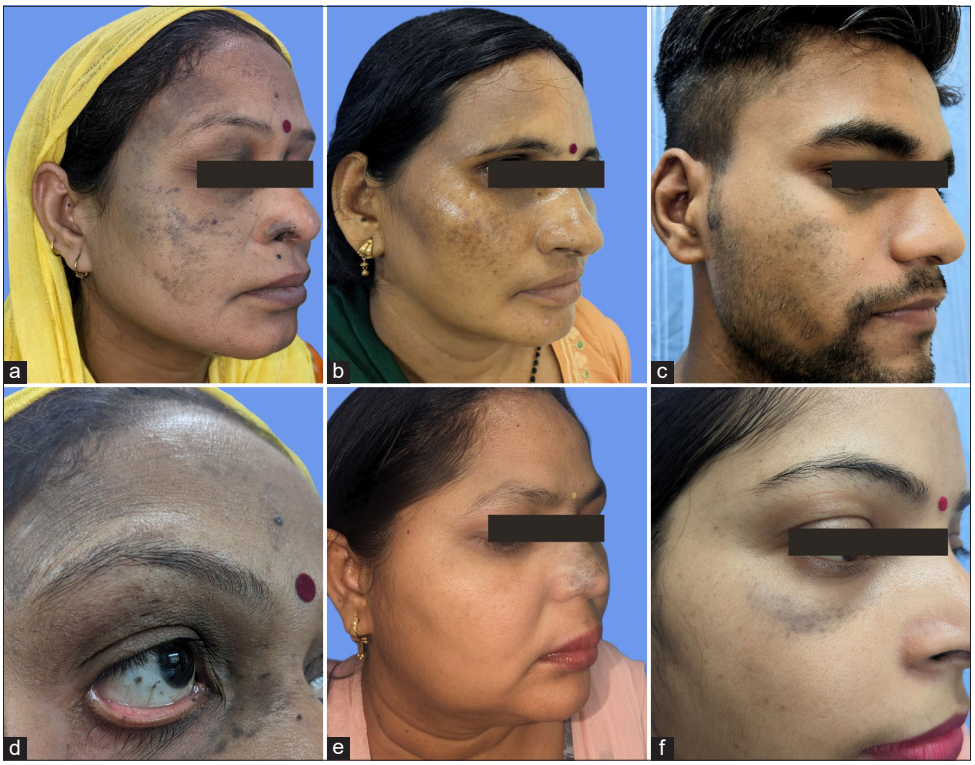

Figure 1: (a-f) Clinical photos of all cases with unilateral bluish-brown discrete to coalescing macules over the face. (d) Scleral pigmentation is shown.

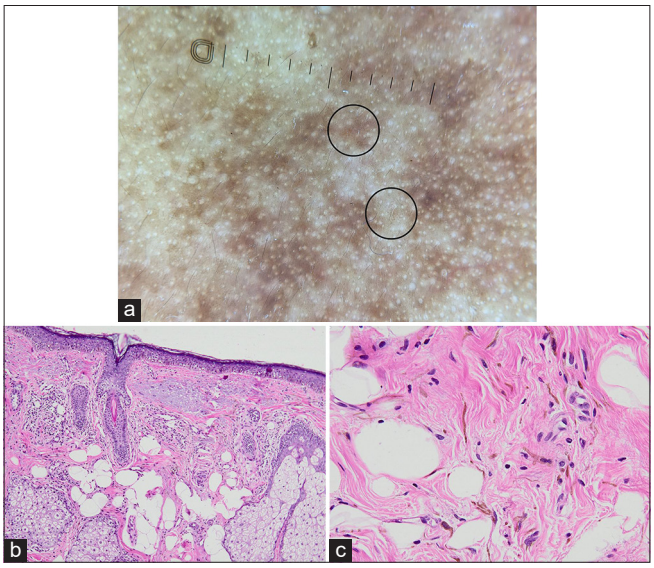

Figure 2: (a) Dermoscopic examination showing brown structureless areas with multiple scattered brown dots (black circles) (DermLite DL5, polarised, contact, ×10). (b) Photomicrograph shows unremarkable epidermis. Underlying dermis shows prominent solar elastosis and moderate perivascular and perifollicular chronic inflammation. (haematoxylin and eosin [H&E]; ×100). (c) Photomicrograph shows a high-power view of the elongated dendritic melanocyte (black arrows) with evenly dispersed melanin granules scattered within collagen bundles in mid-dermis. (H&E; ×400).

dermoscopic and histopathological characteristics of all the cases are presented in Table 1. The patients were diagnosed as having acquired NOO and were subsequently prescribed Q-switched neodymium-doped yttrium–aluminium–garnet (1064 nanometres) laser treatment.

DISCUSSION

Melanocytes are descendants of neural-crest cell derivatives called melanoblasts which originate just after closure of neural tube.[1] The melanocytes subsequently start migrating as early as 2.5 weeks *in utero* and can be demonstrated in the epidermis by 8 weeks.[1] Failure of melanocytes to reach the epidermis or abnormal migration of melanocytes from stratum basale (dropping-off) or from follicular bulbs leads to trapping of melanocytes in the dermis giving rise to dermal melanocytosis.[2] A vast majority of cases of dermal melanocytoses appear in childhood or puberty and are usually diagnosed clinically. However, there exists a rare group of ADM which is clearly adult-onset and frequently misdiagnosed as lichen planus pigmentosus or melasma. The cutoff age for ADM is taken arbitrarily as 25 years based on existing literature. The first studied and well-characterised ADM is Hori's nevus, described by Hori *et al.* in 1984 as blue-brown macules involving forehead, cheeks, temples and eyelids bilaterally of Asian women in the third-fourth

Table 1: Summary of all cases including their dermoscopic and histopathologic characteristics.

Case	Sex	Age	Clinical findings	Scleral Pigmentation	Dermoscopy	Histopathology
1	Female	45 years	Blue grey macules present on right ala of nose extending to nasal bridge.	Present	Blue-grey structureless areas	Melanocytes in a horizontal distribution in mid-dermis.
2	Female	45 years	Blue-grey macules present on right forehead, temple, zygomatic area, cheeks and ala of nose	Present	Blue-grey structureless areas with scattered brown dots	Melanocytes between collagen bundles extending up to mid-dermis
3	Female	46 years	Blue-grey macules present on right malar prominence	Present	Brown-grey structureless areas	Melanocytes present between collagen bundles in mid-dermis
4	Female	45 years	Brown discrete to coalescing macules on cheeks, zygomatic area and forehead	Present	Brown structureless areas with scattered brown dots	Melanocytes in the mid-dermis
5	Male	33 years	Brown to blue discrete to coalescing macules on right cheek and sub-palpebral area	Present	Blue grey structureless areas with scattered brown dots	Melanocytes present between the collagen bundles in the mid dermis.

Table 2: Previous reports on adult-onset NOO from 1991 to 2023.

Year	Authors	Case (s)	Age of onset (average, years)	Ethnicity	Site	Mucosal involvement	Histopathology
1991	Whitemore *et al.*[6]	1	42	Caucasian	Left temple and cheek	Absent	Not available
1994	Lynn *et al.*[7]	1	79	Caucasian	Left cheek, forehead and scalp	Absent	Not available
1999	Lee *et al.*[8]	1	70	Korean	Left periorbital, temple, forehead and scalp	Absent	Melanocytes in upper reticular dermis.
2002	Chang *et al.*[9]	2	40	Korean	Periorbital area, forehead, nose and cheeks	Absent	Melanocytes in upper to mid-dermis
2013	Quenan *et al.*[10]	1	32	Iraqi	Left eyelid, cheek and forehead	Present	Melanocytes in papillary dermis
2019	Khurana *et al.*[5]	6	38	Indian	Unilateral blue-grey macules along V1/V2 distribution	Present in three cases	Melanocytes throughout the dermis extending up to the dermal subcutaneous junction
2019	Irving Llibran *et al.*[11]	1	33	Hispanic	Periorbital and right temporal	Absent	Melanocytes in reticular dermis
2023	Gupta *et al.*[12]	1	34	Indian	Right forehead, cheek and nose	Present	Not performed

NOO: Nevus of Ota

decade. It typically spares the eye and oral mucosae.[3] The next entity under this classification is the acquired unilateral NOO, of which only 14 cases have been reported in the literature previously. Here, the bluish-grey macules present along the distribution of ophthalmic (V1) or maxillary (V2) distribution of the trigeminal nerve unilaterally. The scleral mucosa is frequently involved. The pathogenesis behind ADMs has been poorly studied. One possible mechanism could be the presence of latent dermal melanocytes which are present unnoticed since birth. Later on, melanin synthesising pathway is activated by ultraviolet radiation, local inflammation, sex hormones or unknown ageing stimuli resulting in pigmentation.[4] Another mechanism could be the presence of extracellular sheath around dermal melanocytes which confer it protection and stability.[4] This sheath has been found to be less developed in Mongolian

spots resulting in the regression of pigmentation whereas in NOO, the sheath is thick causing pigmentation to persist.[4]

There exists no difference in the distribution of spindle-shaped melanocytes in the dermis since both acquired unilateral NOO and Hori's nevus show melanocytes in upper and mid-dermis. Likewise in our cases, we found that the dermal melanocytes are present in the mid-dermis in the majority of the cases which correlates with the blue and grey structureless areas on dermoscopy. This is in contrast to a previous study which found that the melanocytes are present throughout the dermis extending to the dermal-subcutaneous junction.[5] Hence, the distribution of melanocytes in the dermis cannot be reliably used to differentiate between Hori's nevus and late-onset unilateral NOO. Table 2 provides a brief summary of existing reports on late-onset unilateral NOO.[6-12]

CONCLUSION

The psychological impact of NOO on patients is immense, which warrants prompt treatment. Due to its rarity in adults, it is crucial for dermatologists to be aware of this condition.

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