

**CASE REPORT**

Nevus of Ota Associated with Nevus of Ito : A Rare Case Report

Noel Sam Thomas, Neha Mariam Joseph

Abstract

Except for the difference in distribution, Nevus of Ito and Nevus of Ota are very similar. In this report, we present the rare simultaneous occurrence of Nevus of Ota and Nevus of Ito in a 21-year-old female patient. Her histopathological examination reveals dermal dendritic melanocytes scattered in the upper portion of the dermis of the lesions. Pulsed Q-switched laser surgery is the choice of treatment for the simultaneous occurrence of Nevus of Ota and Nevus of Ito.

Key Words

Nevus of Ota, Nevus of Ito, Dermal Melanocytosis

Introduction

Nevus of Ito and Nevus of Ota share similar clinical characteristics, with the primary difference being their anatomical distribution. Nevus of Ota, first described by Minor Ota in 1954, is a benign dermal melanosis that primarily affects the cutaneous and ocular regions supplied by branches of the trigeminal nerve.^[1] It is characterized by hyperpigmentation of the skin and mucosal membrane in the distribution of the ophthalmic, maxillary and occasionally, the mandibular divisions of the facial nerve.^[2] The forehead, temples, periorbital area, face, and nose are the most commonly affected areas.^[3] Nevus of Ota often involves pigmentation of the sclera and is associated with significant eye-related complications, including elevated intraocular pressure and the potential development of glaucoma.^[4] Nevus of Ito generally appears at birth and typically affects only one side of the body. It manifests as a bluish-brown mottled pigmentation along the sensory distribution of the posterior supraclavicular and lateral cutaneous branches of the brachial plexus.^[5] The blue-grey patchy pigmentation typically appears on the shoulder, supraclavicular region,

neck, upper arm, scapula, and deltoid area. Nevus of Ito doesn't resolve or lighten with time but may become larger and darker.^[6]

Nevus of Ota affects between 0.014%-0.034% of the Asian population, and up to 0.8% of dermatologic outpatients in Japan; however, there are no reliable incidence or prevalence estimates for the simultaneous occurrence of nevus of Ota and nevus of Ito—globally or in India.^[7,8] Only a handful of independent case reports have documented the simultaneous occurrence of these lesions unilaterally, implying that these are extremely rare. In this report we present the rare simultaneous occurrence of Nevus of Ota and Nevus of Ito.

Case Report

A 21-year-old female patient presented with complaints of bluish grey asymptomatic pigmentation on the left side of her cheek and a brownish pigmentation on her left shoulder. The lesion over the face and shoulder appeared shortly after birth, the hyperpigmentation extended towards subclavicular region and became more pronounced after puberty.

Department of General Medicine, Saveetha Medical College and Hospital, Thandalam, Chennai 602105, Tamil Nadu, India.

Correspondence to: Dr. Noel Sam Thomas, Post Graduate Deptt. of General Medicine, Saveetha Medical College and Hospital, Thandalam, Chennai 602105, Tamil Nadu, India.

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Figure 1: Bluish grey pigmentation on the left cheek.



Figure 2a: Large brownish-grey mottled pigmentation over left shoulder extending towards the ipsilateral arm.

Figure 2b: Mild hyperpigmentation over the left subclavicular region.

On dermatological examination, bluish coloured pigmentation was noted on the left side of the face (Figure 1), large greyish brown mottled pigmentation was noted on the left shoulder which extended towards arm (Figure 2a) and mild hyperpigmentation on left subclavicular region (Figure 2b). General examination, blood parameters, chest radiographs and CT scan of the brain were done, and no abnormalities were found. To evaluate the condition's effect on the eyes, indirect gonioscopy and visual acuity tests were performed, however no abnormalities were found. Additionally, an intraoral examination was performed which showed no abnormality. Histopathology shows numerous dendritic

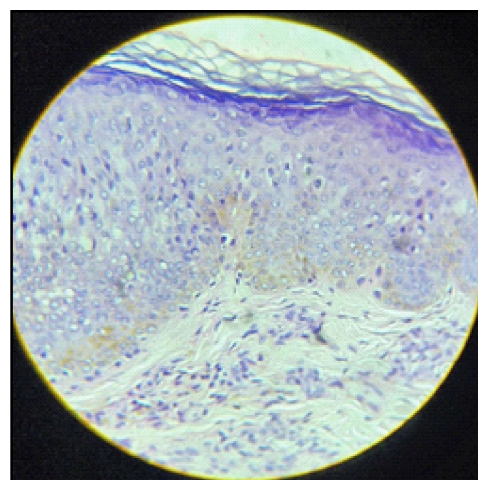


Figure 3: Dermal melanocytes scattered in the upper portion of the dermis of the lesions.

melanocytes dispersed within the upper dermis of the affected skin (Figure 3).

Discussion

Nevus of Ota arises due to the incomplete migration of melanocytes from the neural crest, leading to their entrapment within the dermis rather than reaching the dermo-epidermal junction. The characteristic grey-blue pigmentation is attributed to dermally located melanocytes and the Tyndall effect. This condition is more common among women when compared to men but is rare among Indians. Tanino classified nevus of Ota into four types based on the extent of involvement: Type I (mild), Type II (moderate), Type III (intense), and Type IV (bilateral involvement). In deeper lesions, the Tyndall effect causes the pigmentation to appear blue, whereas superficial lesions tend to exhibit a slate-grey hue, as observed in our case.

Ocular pigmentation in these patients may be associated with a range of eye-related abnormalities, including uveitis, cataract formation, orbital melanoma, retinitis pigmentosa, and asymmetric optic disc cupping not attributable to glaucoma. Additionally, some individuals may exhibit elevated intraocular pressure, which can occur with or without the presence of glaucoma.^[9]

Nevus of Ota is more frequently observed in individuals of Asian descent and those with darker skin phototypes. It typically presents unilaterally, although bilateral cases are occasionally reported. The simultaneous presence of both nevus of Ota and nevus of Ito is extremely rare, with only a limited number of cases documented in the literature.^[10,11]



In the study conducted by *Shanmuga Sekar et al.*^[2], most patients with nevus of Ota presented during their third decade of life, with a female predominance observed. The most common subtype identified was Tanino's type II. Rare presentations noted in their research included bilateral nevus of Ota accompanied by nevus of Ito, as well as cases involving bilateral nevus of Ota with bilateral haemangioma and glaucoma. While the lesions are typically asymptomatic, a lifetime of surveillance is necessary due to the rare, documented incidences of malignant melanoma.^[2]

When treating Ota nevi medically, topical therapy is ineffective. Previous methods of treatment that have been linked to scarring include microsurgery, dermabrasion, and cryotherapy. Q-Switched Nd:YAG laser (QSYL) and the Q-switched ruby laser (QSRL) have made it possible to completely and painlessly remove pigmentation in those with Ota nevus. In the absence of therapy, the skin lesions last a lifetime. In addition, early treatment can reduce the psychological impact of these lesions, which is beneficial to mental health".^[12]

Conclusion

The concurrent occurrence of Nevus of Ota and Nevus of Ito is exceedingly rare, with only a few cases documented in the literature. Its rarity among individuals of Indian origin further underscores the significance of reporting such cases.

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Conflict of Interest: Nil

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