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Case Report

Dentistry Section

Nevus of Ota"- A Rare Pigmentation Disorder with Intraoral Findings

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ABSTRACT

Nevus of Ota is a dermal melanocytosis seen along the distribution of ophthalmic and maxillary divisions of the trigeminal nerve. Only 12 cases so far have been reported in English literature & it is rare in Indian subcontinent. Most of the cases reported are in females & oral cavity is infrequently involved. Here, we report a rare case of unilateral Nevus of Ota in a 56 year old male with oral manifestations.

Keywords: Intra oral pigmentation, Nevi, Oral cavity, Palate, Unliateral

CASE REPORT

A 56-year-old male visited Department of Oral Medicine & Radiology of Vyas Dental College & Hospital, Jodhpur (India) with a chief complaint of decayed tooth in left lower back tooth region of mouth. There was no significant medical history reported. Extra orally, Patient had asymptomatic, speckled, coalescing well-demarcated bluish black hyper pigmented macule on the left mid face involving temporal, frontal, zygoma and the maxillary sinus area [Table/Fig-1]. Bluish pigmentation in sclera of left eye was also observed [Table/Fig-2]. It was present since birth, and family history was noncontributory. Patient also reported that the macules turns dark, deep blue in color in extreme winter and summer seasons. It takes the present color after the weather changes. There was no relevant drug history and no history of photosensitivity. On intraoral examination, blackish discoloration approximately 2 cm in diameter with diffuse margins was evident on the left side of soft palate [Table/Fig-3]. There were also petechiae about two in number present along left retromolar trigone region of mouth. Patient was referred to dermatology for consultation, which did not report any abnormality other than discoloration of face. Auditory examination also did not reveal any discoloration of the auricle and tympanic membrane. A clinical diagnosis of Nevus of Ota was given with the consensus of dermatologist. The patient was not willing for any kind of treatment regarding the pigmentation. Patient was advised to report for regular follow up regarding nevus of Ota. The dental treatment plan formulated for the patient was restoration of 36 & thereafter oral prophylaxis since patient had moderate amount of stains & calculus.

DISCUSSION

Nevus of Ota originally described as nevusfuscocaeruleus ophthalmomaxillaris by Ota and Tanino in 1939 is a dermal melanocytosis most commonly found in Japanese [1,2]. Isolated cases of delayed-onset acquired nevi of Ota that first appear in adults, including in older patients, have been reported3. It is said to be most prevalent in Japan where the incidence among the dermatology outpatients lies between 0.2% to 1%. In Indians it is comparatively rare with the male to female sex ratio being 1:4.8 [4].

Pathophysiology although unconfirmed, yet postulated that Nevus of Ota & other dermal melanocytic disorders such as nevus of ota, blue nevus & mongoloin spots may represent melanocytes that have not migrated completely from neural crest to the epidermis during embryonic stage The two peak ages of onset in early infancy and in early adolescence suggest that hormones are a factor in the development of this condition [5].

After the onset nevus of Ota may slowly & progressively enlarge & darken in color. Its appearance usually remains stable once adulthood is reached. The color & perception of nevus of Ota may fluctuate depending on personal & environmental factors such as fatigue, menstruation , insomnia, and cloudy cold & hot weather conditions [6].

Nevus of Ota often occurs in association with nevus of Ito which is a dermal melanocytic condition affecting the shoulder area. It can also be associated with other cutaneous disorders and ocular disease. Benign cutaneous and leptomeningeal conditions associated with nevus of Ota are phakomatosis pigmentovasculari, nevus flammeus, Sturge-Weber syndrome, Takayasu disease, Klippel-Trenaunay syndrome, and neurofibromatosis [7].

Mishima classified nevus of Ota into three types depending on the extent and distribution of pigmentation [Table/Fig-4]. Thus our case will correspond to Type III classification, as there was involvement of areas innervated by second and third divisions of trigeminal nerve [8].

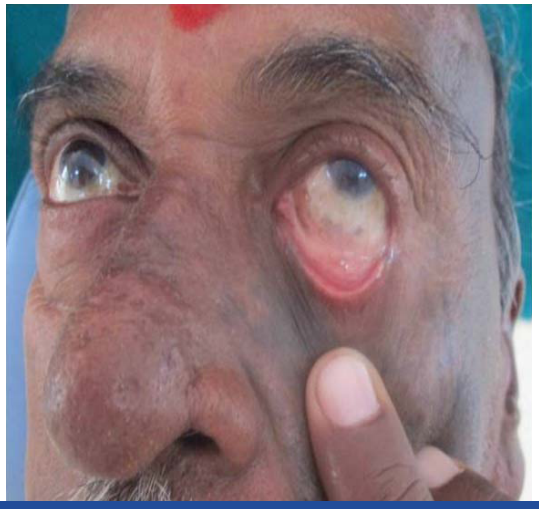
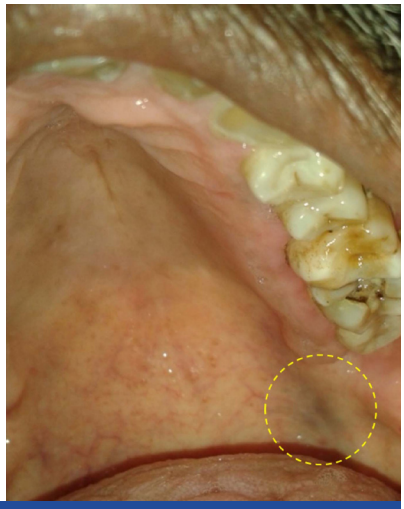
Clinical differential diagnosis for skin lesions of nevus of Ota includes mongolian spot, melasma, blue nevus, and drug-induced hyperpigmentation. Mongolian spot tends to spontaneously resolve by age 3–6 years and typically occurs in lumbosacral areas rarely in face. Melasma is bilateral, associated with pregnancy and is seen without palatal involvement [8].

Drug induced hyperpigmentation is usually acquired after ingestion of drugs like minocycline, amiodarone and gold whereas blue nevus may occur anywhere on skin. Sclera and oral mucosa are not involved in acquired bilateral nevus of Ota like macules (ABNOM) or Sun's nevus [7,8].

Oral melanotic macule can also be misdiagnosed as nevus of Ota. It can also exist in palate but can be distinguished by small in size, and no involvement of sclera. Oral melanotic macule can occur solitary or as a part of syndromes like Peutz-Jeghers syndrome, LEOPARD syndrome, and Laugier-hanziker syndrome. Our patient, however, had no pigmentation of nail, perioral pigmentation, lentigines, and deafness, thus exclusion of above syndromes was made [8].

There is no definitive diagnosis for nevus of Ota. Skin biopsies are required only if clinical changes are suspected of malignant transformation within the involved skin, ocular tissues or mucosal tissues.

Involvement of palatal mucosa occurs infrequently in nevus of Ota. So far, only 11 cases are reported with intraoral involvement with female predominance (64%). Appearance of palatal pigmentation usually blends with oral mucosa and is seen as irregular, ill defined and often mottled patch. Palatal pigmentation can be differentiated

[Table/Fig-1]: Bluish black hyper pigmented macule on the left mid face **[Table/Fig-2]:** Bluish pigmentation in sclera of left eye **[Table/Fig-3]:** Blackish discoloration on the left side of soft palate

Subtypes	Intensity	Pigmentation	Area involved
Type I	Mild	Light brown	Upper and lower eyelids and zygomatic area
Type II	Moderate	Deep slate Gray	Eyelids, zygomatic area, and base of nose
Type III	Intensive	Deep blue to brown	Affecting the first and second divison of trigeminal neuralgia

[Table/Fig-4]: Mishima's Classification of nevus of ota

from blue nevus as blue nevus is slightly raised and is usually less than 1 cm in size. Biopsy is generally not indicated.

The ocular complications associated with nevus of Ota are increased intraocular pressure and glaucoma . Melanoma arising in orbit, iris, ciliary body, and optic nerve in association with nevus of Ota has been documented. Periodic reevaluation with ophthalmologist should also be performed annually.

Topical therapy is of no value in the medical treatment. Previous treatment modalities, including cryotherapy, dermabrasion, and microsurgery, can be associated with scarring. Development of the Q-Switched Nd: YAG laser (QSYL) and the Q-switched ruby laser (QSRL) has enabled complete, scarless elimination of the pigmentation in patients. Without treatment, the skin lesions are permanent.

CONCLUSION

Nevus of Ota with palatal involvement is a rare entity. Dentists should have a thorough knowledge regarding the pigmented lesions as it can indicate varying medical conditions and can lead to future complications if not diagnosed early. Proper follow up and early referral to dermatologist and ophthalmologist is important in diagnosed cases of Nevus of Ota.

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