Case Report

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Nevus of Ota with Intraoral Involvement: Case Report and Review of the Literature

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Abstract

We describe an extraordinary case of a nevus of Ota with intraoral mucosal involvement. We performed a selective review of the English literature and discuss the potential malignant behavior when the oral mucosa is involved.

Keywords: Nevus of ota; Mouth diseases; Hyperpigmentation; Hypermelanosis

Introduction

Nevus of Ota (ocular dermal melanocytosis, congenital melanosis bulbi, nevus fuscoceruleus, ophthalmomaxillaris, oculodermal melanocytosis, oculomucodermal melanocytosis) [1], is a benign pigmentary dermatosis in which melanocytes are found deep within the dermis and sclera [2] due to proliferation and nesting of fusiform melanocytes that follow the first and second branches of the trigeminal nerve [3], resulting in clinical blue pigmentation [4]. It was first described by Hulke in 1860 and later detailed by Ota in 1939 [2,5]. Although its etiology is unknown, it is believed to result from abnormal migration of melanoblastic cells within the primitive neural tube along the trigeminal nerve during embryogenesis. Some studies have revealed that hormonal alterations and exposure to ultraviolet light may have an important role in the pathogenesis of this condition [6].

Nevus of Ota is a rare entity and occurs five times more frequently in women than in men [7], although this datum may be a consequence of an inter consultation bias resulting from aesthetic concerns [2]. Worldwide,

the condition mainly affects patients of Asian and African origin and is uncommon in Caucasians [5,7]. Its exact prevalence among the Mexican population is unknown; however, the disorder was present in 0.7% of patients with facial hyperchromic spots in a dermatologic consultation center [8].

Nevus of Ota is generally characterized by confluent greyish-blue unilateral patches with ill-defined margins, involving preferentially the areas innervated by the first and second divisions of the trigeminal nerve [2,5]. The color of the lesion depends on the location of melanocytes; deeper lesions appear blue due to the Tyndall effect [4]. Lesions may or may not be present at birth and tend to persist through life [9].

According to its distribution, nevus of Ota has been classified as type I or mild, affecting the orbit (I-A), zygomatic (I-B), frontal (I-C) and nasal (I-D) regions; type II or moderate, involving eyelids, periocular, zygomatic, cheek and temporal regions; type III, affecting scalp, forehead, eyebrows and nose; and type IV or bilateral [4,10-12]. It may also be classified according to the

pigmentation and extension of the affected area [13] as type I or mild, in which lesions are light brown and affect the zygomatic region, and both upper and lower eyelids; type II or moderate, characterized by deep slate-grey lesions involving zygomatic area, eyelids and base of the nose; and type III or intense, with lesions that range from deep blue to brown in the regions of the first and second branches of the trigeminal nerve [13].

Nevus of Ota is essentially cutaneous, but often affects the mucosa. Pigmentation can affect the sclera, conjunctiva, cornea, iris, choroid, and less frequently the optic nerve, retrobulbar fat or the orbital periosteum [14]. Melanocytosis may also affect the oral and nasal mucosa, external auditory canal, tympanic membrane, orbital fissures, meninges and the brain [15,16].

The aim of this manuscript is to present an exceptional case of a bilateral nevus of Ota with oral cavity involvement. Additionally, we review the cases published in the English literature, analyzing documents that at least included an abstract in English.

Case report

A 40 year-old Mexican female presented to an Oral Pathology Clinic due to an asymptomatic facial dermatosis present since birth. On skin examination, the patient presented phototype V and diffuse brownish-gray macules on both eyelids and conjunctiva, and left periauricular skin and external ear (Figure 1A), eyelids and conjunctiva (Figure 1B). Intraorally, we observed multiple brown to greyish macules with irregular and diffuse margins affecting the right hemipalate folds and posterior (glandular) zone of hard palate (Figure 1C). No other medical relevant findings were detected.

We made a clinical diagnosis of oculodermal melanocytosis of Ota type IV [10] by location, and type III by intensity. In the differential diagnosis, we considered amalgam tattoo, hypermelanosis associated to systemic diseases, or hyperpigmentation secondary to drugs. Due to the brown-gray discoloration of the oral lesions and the absence of systemic disease or medication intake, we excluded the above diagnoses. The patient did not consent to a biopsy and was later lost to follow-up.

Discussion

The present case was classified as oculodermal melanocytosis of Ota type IV (or bilateral) [10] by location, and type III by intensity; [13,17] only 1-5% of nevi of Ota

[18,19] belong to this type. Intraoral pigmentation in nevus of Ota is extremely rare, and only 45 cases including ours have been described in the English literature (Table 1); being the palate the most frequently involved area [20]. We present a case with palate manifestations and bilateral facial distribution, it is the ninth case reported in the English literature (Table 1) and the first in our country. Nevus of Ota is a well-established entity essentially diagnosed by clinical characteristics and classic morphology. Main differential diagnoses include nevi of Hori and Ito [10,21], drug eruptions, post-inflammatory hyperpigmentation, lichen planus pigmentosus, Sturge-Weber syndrome, *café-au-lait* macules, blue nevi, melasma and actinic lentigo [5]. Intraorally, the differential diagnoses comprise amalgam/metal tattoo, melanocytic nevi, melanosis of multiple causes (including racial, physiologic, endocrine or tobacco-induced post-inflammatory hyperpigmentation), and melanoma.

Figure 1: Clinical findings in a female patient with nevus of Ota. A. Slate-gray patches affecting the ear lobe, helix, and retroauricular skin B. Macules and patches on the eyelids and conjunctiva, concealed by heavy makeup. C. Multiple macules with irregular and diffuse limits involving the hard palate.

Dermal dendritic melanocytic proliferations, including nevus of Ota, are still poorly defined from a dermoscopic point of view. Most of the descriptions are based on the findings of the blue nevi. The most common dermoscopic feature of the latter is a homogeneous bluish to steel-blue pigmentation. Dermoscopy is a routine diagnostic technique that may aid in the diagnosis of nevus of Ota.

On histological examination, nevus of Ota is defined by the presence of dendritic melanocytes dispersed among collagen fibers in the papillary dermis in the absence of stromal scleral reaction [3]. It has been classified into five types according to melanocyte distribution: superficial, superficial dominant, diffuse, deep dominant, and deep

Table 1: English literature review of cases with nevus of Ota and intraoral involvement.

Case/ Reference	Gender/ Age	Intraoral location	Extraoral location	Skin involvement		Other relevant find- ings
				Unilateral	Bilateral	
1/28	♂/16	Buccal mu- cosa	Face and sclera	✓		Melanoma*
2/29	♂/35	Palate & tongue	Cheek, eyelid, chornea		✓	Nevus of Ito. Black pa- tient
3/29	♂/45	Palate	Malar, conjunctivae, nose, chornea, iris, uveal, optic fundus	✓		Nevus of Ito. Black pa- tient
4/30	♂/23	Palate	Ocular, ala nasi, vermillion, forehead, eyelid, temporal, ear		✓	Nevus of Ito
5/31	♂/23	Buccal mu- cosa	Eyelid, sclerae	✓		-
6/32	♀/16	Palate	Sclerae	✓		-
7/33	♀/43	Palate	Sclerae, iris, ala nasi	✓		Sensorineural deafness
8/34	♀/27	Buccal mu- cosa	Ocular, cheek, eyelid	✓		Melanocytoma **
9/35	♀/63	Buccal mu- cosa	Forehead, temporal, eyelids, cheek, conjunctivae	✓		Melanoma**
10/16	♀/59	Palate	Periorbital, sclerae	✓		-
11/36	♀/30	Palate	Ophtalmic and maxilar branch skin area		✓	Klippel-Trenaunay-We- ber syndrome
12/37	♀/30	Palate	Nose, cheek, forehead, sclerae, eye- lids		✓	-
13/7	♀/32	Palate	Sclerae, retina, chornea.	✓		-
14/38	♀/26	Palate & buccal mucosa			✓	Caucasian
15/39	♀/32	Palate	Temporal, nose, cheek, conjunctivae, sclerae.	✓		-
16/39	♀/33	Palate & gingiva	Nose, retroauriculo-temporal, cheek, conjunctivae, sclerae.		✓	-
17-19/4	ND/ND	Palate	ND	ND	ND	-
20/40	♀/3	Palate	Hemifacial	✓		Nevus of Ito. Hereditary
21/40	♀/46	Palate	Periorbital, sclerae.	✓		Nevus of Ito
22/41	♂/21	palate	Forehead, sclerae, conjunctivae	✓		Melanoma**
23/42	♂/18	Palate & gingiva	Forehead, temples, malar, ala nasi, sclerae, conjunctivae	✓		-
24/13	♂/22	Palate	Forehead, temporal, zigomatic, maxi- lar sinus, sclerae	✓		-
25/43	♂/23	Buccal mu- cosa	Malar, periorbital, cheek, sclerae, pin- na of the ear	✓		-
26/44	♀/ND	Palate	Sclerae	✓		Black patient
27/45	♀/36	Palate	Temporal, scalp, forehead, zygomatic arch, periorbital area, sclera, lower palpebra, and radix of the nose	✓		-
28/46	♀/ND	Palate	ND		✓	-
29/18	♀/27	Palate & buccal mucosa		✓		Melanoma*
30/47	♀/30	Palate	Periorbital, sclerae		✓	Klippel-Trenaunay-We- ber syndrome
31/2	♀/25	Buccal mu- cosa	Malar, periorbital.	✓		-
32/48	♀/26	Palate	Forehead, temporal, zigomatic, maxi- lar sinus	✓		-
33/49	♂/22	Palate	Sclerae, ala nasi	✓		-
34/50	♂/24	Palate	Temple, mandibular, malar, sclerae	✓		Bilateral Nevus of Ito
35/27	♀/42	Palate	Forehead, ear, eyelid	✓		Melanoma **
36/17	♂/56	Palate	Temporal, frontal	✓		-
37/23	♂/34	Palate	Eyelid, sclerae	✓		-
38/51	♀/20	Palate & gingiva, tongue	Forehead, sclerae	✓		-
39/26	♂/12	Buccal mu- cosa	Parotid gland, ear	✓		Melanoma*
40/52	♀/20	Buccal mu- cosa	Cheek, ocular	✓		Melanocytoma**
41/53	♂/52	Palate	Periorbital sclera, eyelid	✓		-
42/54	♂/28	Palate	Sclerae, auditory meatus, hemifacial	✓		-
43/54	♀/35	Palate	Hemifacial, sclerae	✓		-
44/55	♀/6	Palate	Forehead, sclerae, temporal	✓		Vitiligo
45/Present case	♀/40	Palate	Ocular, ear		✓	-

*Oral Ota nevus component contiguous to malignant neoplasia; **Oral Ota nevus component not contiguous to malignant neoplasia; ND=No data

[22]; in the later, histopathology is essential to rule-out melanoma [23]. In our case, it was proposed to confirm the diagnosis histologically, but the patient rejected the procedure.

Nevus of Ota is generally asymptomatic and benign, although clinical appearance is a common complaint [2]. Certain conditions have been associated to the presence of nevus of Ota, including Sturge-Weber syndrome, Klippel-Trenaunay syndrome, neurofibromatosis, multiple hemangioma, cerebrosplinal degeneration, glaucoma, and ipsilateral deafness and congenital cataracts [5,13,19,24]. Furthermore, more than 50 cases of melanoma developing on a nevus of Ota have been described [20,25]. Several of these cases occur intracranially, most of them are congenital, and some are accompanied by the presence of hyperpigmented macules in the intraoral mucosa [18,26].

Because of its rarity, epidemiologic studies that describe the malignant transformation of nevi of Ota are lacking. Nevertheless, it seems that malignant transformation is more common in females (male-female ratio 1:7.2); when mucosa is involved (mainly ocular mucosa) [18], and in Caucasians [18,25], amongst who the risk of developing malignant melanoma, although rarely observed [2], is 35 times greater [18] than in other racial groups. Compared with cutaneous and ocular melanoma, oral malignant melanoma is associated with lower rates of 5-year survival and mutations are less common [5]; GNAQ/GNA11 p.Q209L mutation has been found in 77% of primary uveal melanomas, but it is rare in oral mucosal melanoma. Therefore, it is recommended to perform early annual ophthalmologic examination for congenital nevus of Ota with eye involvement, and magnetic resonance imaging screening for all patients beginning at age 25 and in cases with incompletely excised melanocytomas. Interestingly, concurrent development of malignancy has been described in eight subjects (17.8%) with intraoral nevus of Ota (Table 1). In all of them, malignancy was ipsilateral, two cases adjacent to the nevi and one developed from the parotid gland [26]. Although cancer rarely develops within nevi of Ota, the percentage of malignant transformation observed when the oral mucosa is involved should be considered when evaluating subjects. This risk should not be underestimated, and histopathological analysis is advised to confirm the diagnosis and rule out melanoma [18,25]. In the presence of malignant melanoma, the excision of the entire intraoral lesion (nevi of Ota) is recommended. In our case, the presence of melanosis in

the palate, female gender, bilaterality and ocular lesions supported the need to perform a biopsy or completely remove the lesion from the palatine mucosa.

In summary, we describe an unusual clinical presentation of a nevus of Ota. Although the possibility of malignant transformation from a nevus of Ota is low, the risk should be considered [27] and biannual follow-up is recommended to monitor sudden changes in color, ulceration or paresthesia [13]. In the context of nevus of Ota with oral involvement, it is important to consider that 17.8% of the cases can develop malignant tumors; thus, histopathology is essential to confirm the condition and to discard malignancy, notwithstanding its low frequency [23].

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Conflict of interest

None reported

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References

1. Redkar NN, Rawat KJ, Warriar S, Jena A. Nevus of Ota. J Assoc Physicians India 2016; 64:70. PMID: http://www.japi.org/april_2016/10_pc_nevus_of_ota.html
2. Mohan RP, Verma S, Singh AK, Singh U. 'Nevi of Ota: the unusual birthmarks': a case review. BMJ Case Rep 2013; PMID: <https://www.ncbi.nlm.nih.gov/pubmed/23456162>
3. Chan HH, Kono T. Nevus of Ota: Clinical aspects and management. Skinmed 2003; 2:89-96. PMID: <https://www.ncbi.nlm.nih.gov/pubmed/14673306>
4. Sekar S, Kuruvila M, Pai HS. Nevus of Ota: a series of 15 cases. Indian J Dermatol Venereol Leprol 2008; 74:125-127. PMID: <https://www.ncbi.nlm.nih.gov/pubmed/18388370>
5. Alvarez-Cuesta CC, Raya-Aguado C, Vazquez-Lopez F, Garcia PB, Perez-Oliva N. Nevus of Ota associated with ipsilateral deafness. J Am Acad Dermatol 2002; 47:S257-259. PMID: <https://www.ncbi.nlm.nih.gov/m/pubmed/12399743/>
6. Wang BQ, Shen ZY, Fei Y, Li H, Liu JH, Xu H, et al. A population-based study of acquired bilateral nevus-

- of-Ota-like macules in Shanghai, China. *J Invest Dermatol* 2011; 131:358–362. PMID: <https://www.ncbi.nlm.nih.gov/pubmed/20882042>
7. Kannan SK. Oculodermal melanocytosis--Nevus of Ota with palatal pigmentation. *Indian J Dent Res* 2003; 14:230–233. PMID: <https://www.ncbi.nlm.nih.gov/pubmed/15328990>
 8. Arellano-Mendoza MI, Tirado-Sánchez A, Mercadillo-Pérez P, León-Dorantes G, Ponce-Olivera RM. Motivo de consulta: manchas hiperocrómicas en la cara. *Dermatol Rev Mex* 2011; 55:180-184. <http://www.medigraphic.com/pdfs/derrevmex/rmd-2011/rmd114c.pdf>
 9. Mosher DB, Fitzpatrick TB, Ortonne JP, Hori Y. Hypomelanoses and hypermelanoses. In: Freedberg IM, Eisen AZ, Wolff K, Austen KF, Goldsmith LA, Katz SI, Fitzpatrick TB, eds. *Dermatology in general medicine*. New York, NY: McGraw-Hill; 1999;1:945-1017.
 10. Tanino H. Naevus fusco-caeruleus phtharmomaxillaris (Ota). *Jpn J Dermatol* 1939; 46:107–111.
 11. Huang WH, Wang HW, Sun QN, Jin HZ, Liu YH, Ma DL, et al. A new classification of nevus of Ota. *Chin Med J (Engl)* 2013; 126:3910-3914. PMID: <https://www.ncbi.nlm.nih.gov/pubmed/24157155>
 12. Patzwahl R, Landau K, Kollias SS. Atypical midface tumor complicating nevus of Ota. *AJNR Am J Neuroradiol* 2005; 26:2117–2121. PMID: <https://www.ncbi.nlm.nih.gov/pubmed/16155168>
 13. Sharma G, Nagpal A. Nevus of Ota with rare palatal involvement: a case report with emphasis on differential diagnosis. *Case Rep Dent* 2011; 2011:670679. PMID: <https://www.ncbi.nlm.nih.gov/pubmed/22567445>
 14. Patel BC, Egan CA, Lucius RW, Gerwels JW, Mamalis N, Anderson RL. Cutaneous malignant melanoma and oculodermalmelanocytosis (nevus of Ota): report of a case and review of the literature. *J Am Acad Dermatol* 1998; 38:862–865. PMID: <https://www.ncbi.nlm.nih.gov/pubmed/9591805>
 15. Wilcox JC. Melanomatosis of the Skin and Central Nervous System in infants: A Congenital Neurocutaneous Syndrome. *Am J Dis Child* 1939; 57:391–400. PMID: <https://jamanetwork.com/journals/jamapediatrics/article-abstract/1178422>
 16. Page DG, Svirsky JA, Kaugars GE. Nevus of Ota with associated palatal involvement. *Oral Surg Oral Med Oral Pathol* 1985; 59:282–284. PMID: <https://www.ncbi.nlm.nih.gov/pubmed/3856821>
 17. Solanki J, Gupta S, Sharma N, Singh M, Bhateja S. Nevus of Ota"- a rare pigmentation disorder with intraoral findings. *J Clin Diagn Res* 2014; 8:49-50. PMID: <https://www.ncbi.nlm.nih.gov/pubmed/25302286>
 18. Hassan ML, Zambrano R, Vera ME, Schroh RG, Binda M, Rebadan A. Nevo de Ota y melanoma Comunicación de un caso y consideraciones. *Argent Dermatol* 2012; 62:49-55. <http://www.archivosdermato.org.ar/Uploads/049%20Nevo%20de%20Ota%20y%20Melanoma.pdf>
 19. Yang Y, Guo X, Xu J, Ye Y, Liu X, Yu M. Phakomatosis pigmento vascularis associated with Sturge-Weber syndrome, Ota nevus, and congenital glaucoma. *Medicine* 2015; 94: e1025. PMID: <https://www.ncbi.nlm.nih.gov/pubmed/26131807>
 20. Alam M, Arndt KA, Dover JS. Laser treatment of nevus of Ota. *Dermatologic Therapy* 2001; 14:55–59. PMID: <https://onlinelibrary.wiley.com/doi/pdf/10.1046/j.1529-8019.2001.014001055.x>
 21. Lam YM. Review on acquired bilateral nevus of Ota-like macules. *Hong Kong Dermatol Venereol Bull* 2001; 9:60-64. <https://pdfs.semanticscholar.org/f0e8/32c6633a24bb19fa4661dd996afa0a7ac0c4.pdf>
 22. Hirayama T, Suzuki T. A new classification of Ota's nevus based on histopathological features. *Dermatologica* 1991; 183:169-172. PMID: <https://www.ncbi.nlm.nih.gov/pubmed/1743382>
 23. Sehgal VN, Syed NH, Aggarwal A, Sharma S, Sehgal S. Nevus of Ota / oculodermal melanocytosis: a rare report of an oral mucosal lesion involving the hard palate. *Cutis* 2015; 96:10-12. PMID: <https://www.ncbi.nlm.nih.gov/pubmed/26682294>
 24. Teekhasaene C, Ritch R, Rutnin U, Leelawongs N. Glaucoma in oculodermal melanocytosis. *Ophthalmology* 1990; 97:562-570. PMID: <https://www.ncbi.nlm.nih.gov/pubmed/2342801>
 25. Buntinx-krieg T, Ouyang J, Cartwright M. An orbital malignant melanoma arising in cellular blue nevus in a patient with nevus of Ota. *Cureus* 2016; 8:e698. PMID: <https://www.ncbi.nlm.nih.gov/pubmed/27699140>
 26. Lindsey SF, Sanchez MI, Elgart GW, Milikowski C, Civantos FJ, Goldberg J, Grichnik JM. Malignant melanoma from a nevus of Ota in a pediatric patient with fatal outcome. *J Am Acad Dermatol* 2013; 69:195-197. PMID: <https://www.ncbi.nlm.nih.gov/pubmed/24034399>
 27. Nitta K, Kashima T, Mayuzumi H, Akiyama H, Miyanaga T, Hirato J, Kishi S. Animal-type malignant melanoma associated with nevus of Ota in the orbit

- of a Japanese woman: a case report. *Melanoma Res* 2014; 24:286-289. PMID: <https://www.ncbi.nlm.nih.gov/pubmed/24686419>
28. Dorsey CS, Montgomery H. Blue nevus and its distinction from Mongolian spot and the nevus of Ota. *J Invest Dermatol* 1954; 22:225-236. PMID: <https://www.ncbi.nlm.nih.gov/pubmed/13130904>
 29. Mishima Y, Mevorah B. Nevus Ota and nevus Ito in American Negroes. *J Invest Dermatol* 1961; 36:133-154. PMID: <https://www.ncbi.nlm.nih.gov/pubmed/13771275>
 30. Hidano A, Kajima H, Endo Y. Bilateral nevus Ota associated with nevus Ito. A case of pigmentation on the lips. *Arch Dermatol* 1965; 91:357-359. PMID: <https://www.ncbi.nlm.nih.gov/pubmed/14275500>
 31. Estima CA, Carneiro SR. Ota naevus: presentation of a case. *Br J Plast Surg*. 1972; 25(1):49-52. <https://europepmc.org/abstract/med/5008783>
 32. Bhattacharya SK, Girgla HS, Singh G. Nevus of Ota. *Int J Dermatol*. 12(6):344-7. <https://onlinelibrary.wiley.com/doi/abs/10.1111/j.1365-4362.1973.tb00218>.
 33. Reed WB, Sugarman GI. Unilateral nevus of Ota with sensorineural deafness. *Arch Dermatol*. 1974; 109(6):881-3. PMID: <https://www.ncbi.nlm.nih.gov/pubmed/4830102>
 34. Yeschua R, Wexler MR, Neuman Z. The nevus of Ota: Case Report. *Plastic Reconstr Surg*. 1975; 55(2):229. https://journals.lww.com/plasreconsurg/Citation/1975/02000/THE_NEVUS_OF_OTA__Case_Report_.33.aspx
 35. Haim T, Meyer E, Kerner H, Zonis S. Oculodermal melanocytosis (nevus of Ota) and orbital malignant melanoma. *Ann Ophthalmol*. 1982; 14(12):1132-6. PMID: <https://www.ncbi.nlm.nih.gov/pubmed/7165231>
 36. Talwar S, Jha PK, Suresh MS, Panvelkar VVN. Bilateral nevus of Ota with Klippel-Trenaunay-Weber syndrome. *Med J Armed Forces India* 1992; 48:231-233. PMID: <http://medind.nic.in/imvw/imvw18223.html>
 37. Rath SK. Bilateral nevus of ota with oral mucosal involvement. *Indian J Dermatol Venereol Leprol*. 2002; 68(2):104. PMID: <https://www.ncbi.nlm.nih.gov/pubmed/17656896>
 38. Turnbull JR, Assaf C, Zouboulis C, Tebbe B. Bilateral naevus of Ota: a rare manifestation in a Caucasian. *J Eur Acad Dermatol Venereol*. 2004; 18(3):353-5. PMID: <https://www.ncbi.nlm.nih.gov/pubmed/15096155>
 39. Parihar APS, Bagewadi A, Keluskar V, Shetti A. Nevus of Ota Involving Palate: Case Reports and Review. *JIAOMR*. 2007; 19(3):441. <http://www.jiaomr.in/article.asp?issn=0972-1363;year=2007;volume=19;issue=3;page=441;epage=445;aulast=Parihar;type=0>
 40. Agero ALC, Lahmar JJ, Holzborn RM, Martin LK, Freckmann M-L, Murrell DF. Naevus of Ota presenting in two generations: a mother and daughter. *J Eur Acad Dermatol Venereol*. 2009; 23(1):102-4. PMID: <https://www.ncbi.nlm.nih.gov/pubmed/18422534>
 41. Chandak R, Degwekar S, Chandak M, Bhowte R, Rawlani S. Rare case report on nevus of Ota. *J Kor Dent Sci* 2010; 3:43-47.
 42. Cronemberger S, Calixto N, Freitas HL. Nevus of Ota: clinical-ophthalmological findings. *Revista Brasileira de Oftalmologia*. 2011; 70(5):278-83. http://www.scielo.br/scielo.php?script=sci_arttext&pid=S0034-72802011000500002
 43. Azar M, Kazemi F, Bahrami E, Hejazian E, Fereshtehnejad S-M, Ahmadi A, et al. Meningeal melanomas associated with transforming Ota nevus to malignant melanoma: a case report. *Med Islam Repub Iran (MJIRI)*. 2010; 24(3):163-8. http://mjiri.iums.ac.ir/browse.php?a_id=193&sid=1&slc_lang=en
 44. Shetty SR, Subhas BG, Rao KA, Castellino R. Nevus of Ota with buccal mucosal pigmentation: a rare case. *Dent Res J (Isfahan)*. 2011; 8(1):52-5 PMID: <https://www.ncbi.nlm.nih.gov/pmc/articles/PMC3177382/>.
 45. Guledgud MV, Patil K, Srivathsa SH, Malleshi SN. Report of rare palatal expression of Nevus of Ota with amendment of Tanino's classification. *Indian J Dent Res*. 2011; 22(6):850-2. PMID: <https://www.ncbi.nlm.nih.gov/pubmed/22484884>
 46. Alshami M, Bawazir MA, Atwan AA. Nevus of Ota: morphological patterns and distribution in 47 Yemeni cases. *J Eur Acad Dermatol Venereol*. 2012; 26(11):1360-3. PMID: <https://www.ncbi.nlm.nih.gov/pubmed/21967654>
 47. Garg A, Gupta LK, Khare AK, Kuldeep CM, Mittal A, Mehta S. Phacomatosis cesioflammea with Klippel Trenaunay syndrome: A rare association. *Indian Dermatol Online J*. 2013; 4(3):216-8. PMID: <https://www.ncbi.nlm.nih.gov/pubmed/23984239>
 48. Prashanth P, Srikanth D, Venkatesh E, Kumar M. The case of a rare nevus with palatal, facial, and scleral pigmentation. *Univ Res J Dent*. 2013; 3:131. https://www.researchgate.net/publication/272895680_The_case_of_a_rare_nevus_with_palatal_facial_and_scleral_pigmentation

49. Srivathsa SH. Nevus of Ota: presentation of a case with oral pigmentation and codicil to Tanino's classification. 2013; 31:24. <https://www.dermatology.org.my/pdf/journal%202013%20B2.pdf>
50. Mukhopadhyay AK. Unilateral nevus of Ota with bilateral nevus of Ito and palatal lesion: a case report with proposed clinical modification of Tanino's classification. Indian J Dermatol. 2013; 58(4):286-9. PMID: <https://www.ncbi.nlm.nih.gov/pubmed/23918999>
51. Shivhare P, Lata S, Yadav M, Haidry N, Patil ST. A nevus of Ota with intraoral involvement: a rare case report. Int J Oral Health Med Res 2015; 2:76-79. <http://www.ijohmr.com/upload/Nevus%20of%20OTA-%20A%20rare%20entity.pdf>
52. Chen H, Liu W, Zhang S, Xu J, Hui X. Cerebellar meningeal melanocytoma associated with nevus of Ota: An extremely rare case. Neurology. 2015; 85(6):555-6. PMID: <https://www.ncbi.nlm.nih.gov/pubmed/26259856>
53. Tomov GT, Mutafchieva MZ, Nikolov NV. Oral Manifestation of Ota's Nevus. A Case Report with Emphasis on Differential Diagnosis. Oral Surg, Oral Med, Oral Pathol Oral Radiol. 2015; 119(3):e164. [https://www.oooojournal.net/article/S2212-4403\(14\)00973-0/abstract](https://www.oooojournal.net/article/S2212-4403(14)00973-0/abstract)
54. Preethika GB, Anuja J, Roopashri RK, Devika S. Oculodermal melanocytosis: a case series. Sch J Dent Sci 2016; 5:9-10. <http://saspjournals.com/wp-content/uploads/2016/02/SJDS-318-11.pdf>
55. Yarramachu S, Bijina KD, Samayam A. Nevus of Ota with overlying vitiligo a case report. Indian J Med Case Rep 2016; 5:9-10. <http://www.cibtech.org/J-Medical-Case-Reports/PUBLICATIONS/2016/VOL-5-NO-3/03-JCR-014-SWETHA-NEVUS-REPORT.pdf>

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