

Congenital Lingual Melanotic Macule Rare entity in infants

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الطاخة الخلقية الصبغية في اللسان
مرض نادر في الأطفال الرضع

جيتان كومار، ديپا شارما، أكاش بانديتا، سویتا شاستري

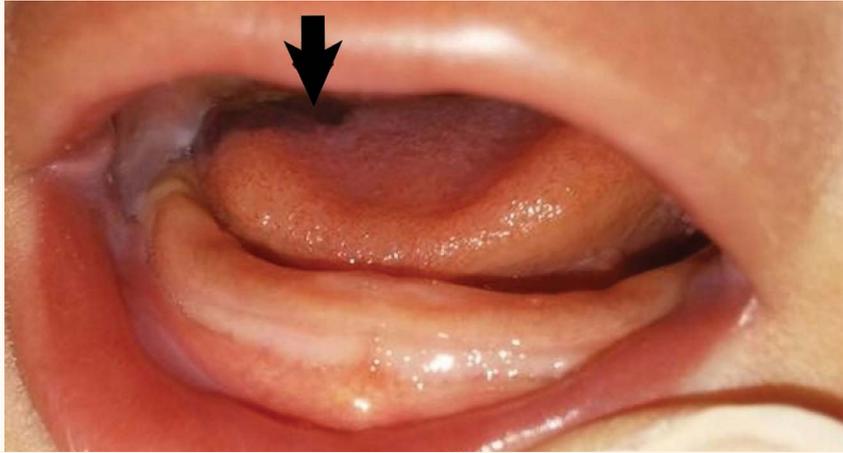


Figure 1: Five-month-old male infant with a solitary black congenital lingual melanotic macule measuring 6 x 6 mm on the right lateral aspect of the surface of the tongue (arrow).

A FIVE-MONTH-OLD MALE INFANT PRESENTED to the Department of Pediatrics at the Madras Institute of Orthopaedics & Traumatology in Chennai, India, in June 2014 with a pigmented lesion on the tongue that had been observed since birth. The baby was healthy, accepting feeds well and was not taking any medication. There was no associated family history of melanomas, polyposis or mucosal pigmentation. The mother reported that the size of the lesion had been increasing gradually since birth. An oral cavity examination revealed a solitary 6 x 6 mm black macule on the right lateral aspect of the surface of the tongue [Figure 1]. There were no other pigmented lesions found anywhere else on the infant's body. A dermatological opinion was sought and a diagnosis of a congenital lingual melanotic macule was made. A biopsy was recommended; however, the family moved away and the patient was lost to follow-up.

Comment

Hyperpigmentation of the oral cavity is usually seen after the second decade of life and can be caused by the intake of certain drugs, smoking or by carcinomatous changes.¹ However, it is very rarely seen in the neonatal and infantile period.² In adults, congenital oral and labial melanotic macules are among the most common cause of hyperpigmented lesions of the oral mucosa.² There are very few case reports of congenital lingual melanotic macules presenting in newborns and infants.³ These macules are now recognised as a unique and benign cause of hyperpigmentation of the tongue.²

Congenital oral and labial melanotic macules are the most common causes of hyperpigmented lesions of the oral mucosa in the elderly population; however, they are rarely observed in the neonatal period.² Oral melanotic macules usually present at a median age of 40

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years, whereas labial melanotic macules usually present at a median age of 27.5 years.⁴ The most common localisation of the macule is a single lesion at the vermillion border of the lower lip near the midline, with the size of the lesion rarely exceeding 5 mm in diameter.⁴

Congenital lingual melanotic macules usually have benign histological features; however, biopsies are recommended to ascertain that the lesion is not malignant. A biopsy of the lesion is characterised by excess deposition of melanin in the basal cell layer and *lamina propria* of the skin.³ Histopathological examination of the lesion shows an increase in melanin pigmentation of the basal epidermal layer and hyperkeratosis of a varying nature with an abundance of subepidermal pigment-laden macrophages.³ There is usually a normal number of melanocyte cells; junctional nests of melanocytes and atypical cells are also noted. Congenital lingual melanotic macules are differentiated from melanocytic naevi and melanomas by the lack of melanocyte nests and by negative melanosome antibody staining (homatropine methyle bromide-45), respectively.³

The differential diagnosis for congenital lingual melanotic macules includes physiological melanin pigmentation; drug-associated pigmentation (e.g. from chloroquine); toxin-associated pigmentation; pigmented fungiform papillae; Laugier-Hunziker syndrome; pigmented naevi; malignant melanomas; smoker's melanosis; amalgam tattoos; post-inflammatory pigmentation; Addison's disease; and Peutz-Jeghers syndrome.⁵ A detailed patient history and thorough physical examination of the lesion are needed to rule out other possible causes of pigmentation of the oral cavity, such as smoking and the intake of certain drugs. Toxin tests and a biopsy should be performed to rule out conditions like malignant or metastatic melanomas, which can also present in a similar manner. Three criteria need to be fulfilled before a clinical diagnosis of congenital lingual

melanotic macule can be made: (1) the presence of single or multiple melanotic lesions on the tongue; (2) the presence of the lesion since birth and which continues to grow in size; and (3) no family history of any disease known to cause mucosal pigmentation.⁴

Patients with congenital lingual melanotic macules need to be followed up regularly to note any changes in the size, shape or colour of the lesion. Long-term outcomes of patients with congenital lingual melanotic macules are not known, as fewer than ten cases have been reported and the cause of origin of the lesion remains uncertain.⁶ There is no specific treatment for oral melanotic macules, although many researchers recommend complete excision and histological examination of the lesion.⁷

References

1. Ho KK, Dervan P, O'Loughlin S, Powell FC. Labial melanotic macule: A clinical, histopathologic, and ultrastructural study. *J Am Acad Dermatol* 1993; 28:33–9. doi: 10.1016/0190-9622(93)70005-e.
2. Marque M, Vabres P, Prigent F, Guillot B, Bessis D. [Congenital melanotic macules of the tongue.] *Ann Dermatol Venerol* 2008; 135:567–70. doi: 10.1016/j.annder.2008.07.012.
3. Menni S, Boccardi D. Melanotic macules of the tongue in a newborn. *J Am Acad Dermatol* 2001; 44:1048–9. doi: 10.1067/mjd.2001.113687.
4. Dohil MA, Billman G, Pransky S, Eichenfield LF. The congenital lingual melanotic macule. *Arch Dermatol* 2003; 139:767–70. doi: 10.1001/archderm.139.6.767.
5. Eisen D, Voorhees JJ. Oral melanoma and other pigmented lesions of the oral cavity. *J Am Acad Dermatol* 1991; 24:527–37. doi: 10.1016/0190-9622(91)70077-F.
6. Rapini RP, Golitz LE, Greer RO Jr, Krekorian EA, Poulson T. Primary malignant melanoma of the oral cavity: A review of 177 cases. *Cancer* 1985; 55:1543–51. doi: 10.1002/1097-0142(19850401)55:7<1543::AID-CNCR2820550722>3.0.CO;2-F.
7. Shen ZY, Liu W, Bao ZX, Zhou ZT, Wang LZ. Oral melanotic macule and primary oral malignant melanoma: Epidemiology, location involved, and clinical implications. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2011; 112:e21–5. doi: 10.1016/j.tripleo.2011.02.040.