Journal of Dermatological Research

Online Submissions: http://www.ghrnet.org/index./jdr/
doi:10.17554/j.issn.2413-8223.2019.04.46-1

J. of Dermatological Res. 2019 December; 4(2): 167-169
ISSN 2413-8223

TOPIC HIGHLIGHT

Congenital Melanotic Lingual Macules

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Conflict-of-interest statement: The author(s) declare(s) that there is no conflict of interest regarding the publication of this paper.

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Received: July 30, 2019
Revised: August 6, 2019
Accepted: September 1, 2019
Published online: December 19, 2019

ABSTRACT

We here present five cases of congenital melanotic lingual macules seen in our hospital. All the patients were healthy and normally thriving. A close follow-up showed that lesions were non-evolutive and allowed us to exclude malignancies. Pigmented oral lesions may show great variability in presentation, hue, number, size, and location in the oral cavity. Congenital, stable lesions generally raise less concern than acquired and/or rapidly evolving spots. As for tongue lesions, an accurate patient history and thorough head-to-toe physical examination in search for other lesions or stigmata of known syndromes is -in most cases- sufficient to exclude malignancies. Lesion excision should therefore not represent the first choice if a close clinical follow up can be established.

Key words: Lingual macules; Pediatrics; Dermatology

Silecchia V, Valerio E, Berti I, Morandi F, Cutrone M, Grimalt R. Congenital Melanotic Lingual Macules. *Journal of Dermatological Research* 2019; **4(2)**: 167-169 Available from: URL: http //www.ghrnet.org/index.php/jdr/article/view/2727

CASE SERIES

Patient 1

A male Italian newborn was referred to us at day of life 11 for four congenital pigmented spots on the upper left side of the tongue. A whitish patch was present all around the pigmented lesions, and contralaterally (Figure 1A), where no melanotic spots were present. The baby was asymptomatic and fed well.

Patient 2

A male Bengalese newborn presented with a pigmented brown streak along the median line of the tongue. The lesion was plain and not painful. He received two weeks of topical antimycotic, without any change in pigmentation. At 4 weeks follow-up visit the lesion was unchanged (Figure 1B).

Patient 3

A 2-month Italian toddler exhibited since birth several black macules on the upper side of the tongue. Surrounding white discoloration was interpreted as a possible mycosis and therefore treated with one week of topical antifungal therapy associated with daily rubbing of lingual surface, without any change in the pigmented spots (Figure 1C).

Patient 4

We evaluated a 1-year-old Bengalese boy for cough and fever in the

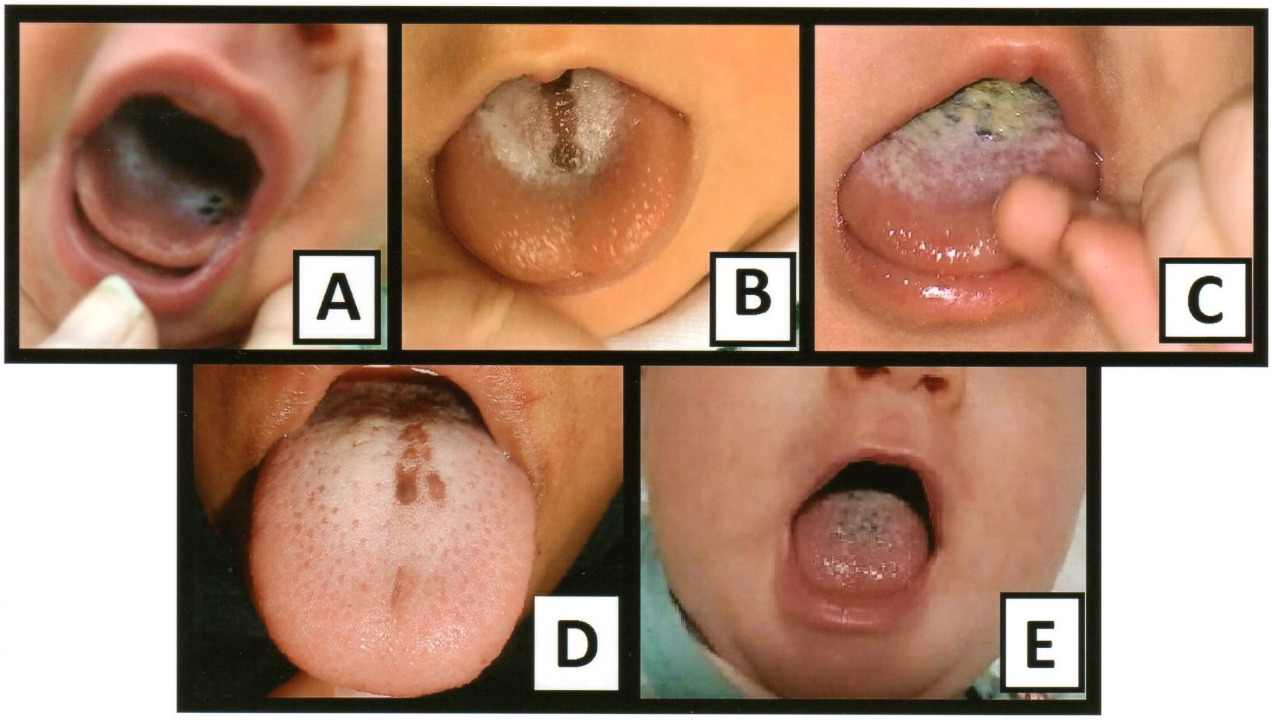

Figure 1 A, congenital pigmented tongue spots in a 11-day male Italian newborn; B, congenital pigmented brown streak along the median line of the tongue in a 4-week male Bengalese newborn; C, black macules on the upper side of the tongue of a 2-month Italian toddler, unchanged after one week of topical antifungal therapy; D, brown spots along the median line of the tongue of a 1-year-old Bengalese boy, reported as present since birth without any growth or modification; E, pigmented macules of the tongue, unchanged from birth, in a 10-week female Italian newborn.

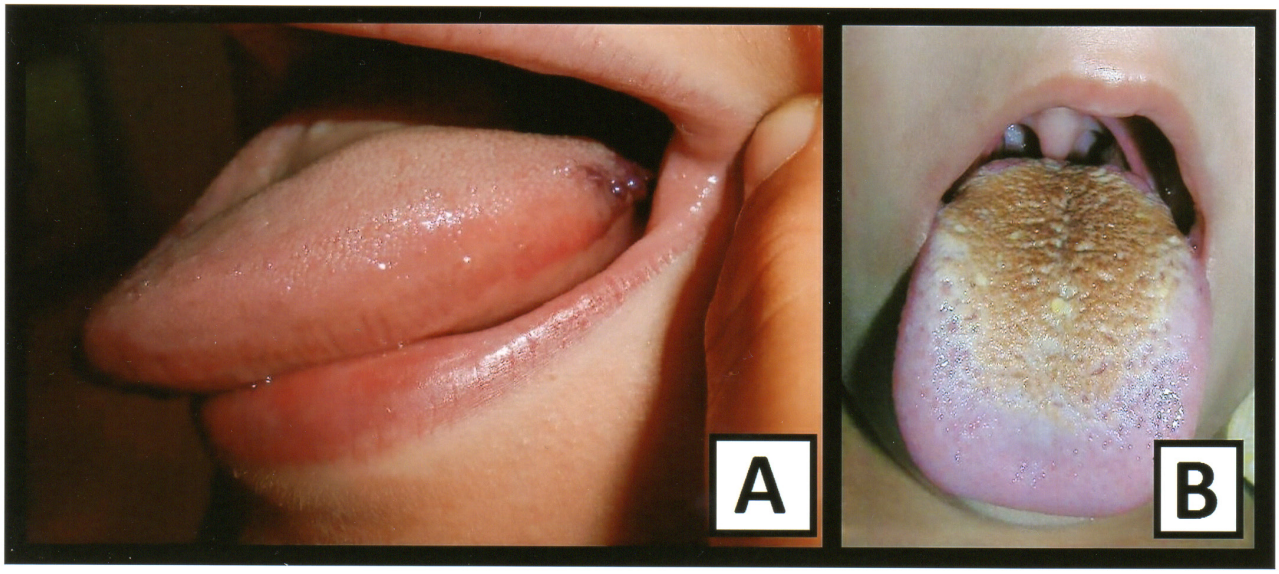

Figure 2 Two possible differential diagnoses of congenital melanotic lingual macules. A, microcystic lymphangioma (usually presenting as vescicular, trasparent "frog eggs-like" lesions, sometimes blackened due to traumatic hemorrhage); B, lingua nigra villosa or black hairy tongue, an acquired condition usually occurring after prolonged antibiotic therapy.

emergency. Incidentally, asymptomatic brown spots along the median line of the tongue were detected (Figure 1D). Parents reported that those lesions were present since birth, and never grew nor modified. The boy was otherwise healthy and thriving.

Patient 5

Pigmented macules of the tongue (Figure 1E) were noticed since birth in a 10-week female Italian newborn. She received two weeks of topical antimycotics with no change in the lesions. The baby was asymptomatic and normally thriving.

DISCUSSION

Multiple conditions may be accountable for focal hyperpigmentation of the oral mucosa: among others, exposure to drugs/toxics; post-traumatic submucosal hemorrhages; small hemangiomas or microcystic lymphangioma (usually presenting as vescicular, trasparent "frog eggs-like" lesions, sometimes blackened due to traumatic hemorrhage, as in the patient of Figure 2A); post-radiotherapy pigmentation[1]; pigmented fungiform papillae; lingua nigra villosa or black hairy tongue (an acquired condition usually

occurring after prolonged antibiotic therapy, depicted in Figure 2B); Laugier-Hunziker syndrome; Peutz-Jeghers syndrome (in which lesions are usually absent at birth and are associated to perioral lesions, often present also in other family members); pigmented nevi[2] or functional melanocytic hamartoma (characterized by a local melanin hyperproduction)[1]; neurofibromatosis (which can be reasonably ruled out on the basis of absence of other lesions)[3].

Finally, the most fearsome differential is melanoma; its extreme rarity in the first years of life and the absolute exceptionality of presentation as a pigmented lesion of the tongue warrant a careful follow up as a sufficient initial measure; in still doubtful cases, the negativity of immunomarkers for HMB-45 on the excised lesion allows to rule out the diagnosis of melanoma and/or melanocytic nevus[1].

None of the lesions presented in our case series changed over time at repeat follow-up controls, and neither was associated to traumatic events or previous treatments; finally, no familiar history of similar lesions was evident in any of the presented patients. These characteristics are compatible with the diagnosis of congenital melanotic lingual macules (CMLM).

CMLM represent a common variant of congenital oral macules; these are benign lesions located on the gingival or buccal mucosa, palate, and/or tongue. They generally appear isolated and well circumscribed, of black, brownish or blue hue; their usual size is often under 10 (frequently 3 or 4) millimeters[3].

CONCLUSIONS AND FINAL REMARKS

Pigmented oral lesions may show great variability in presentation, hue, number, size, and location in the oral cavity. Congenital, stable lesions generally raise less concern than acquired and/or rapidly evolving spots. As for tongue lesions, an accurate patient history and thorough head-to-toe physical examination in search for other lesions or stigmata of known syndromes is -in most cases- sufficient to exclude malignancies. Lesion excision should therefore not represent the first choice if a close clinical follow up can be established.

Contributorship Statement

Valeria Silecchia wrote the first draft of the manuscript; Irene Berti, Francesco Morandi, Mario Cutrone, and Ramon Grimalt contributed to the critical revision of the manuscript and obtained the iconographic documentation.

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