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CASE REPORT

Idiopathic Lenticular Pigmentation – A Case Report with Differential Diagnosis

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ABSTRACT

Pigmentation of oral mucosa is very common which can present in different clinical patterns and colour depending on the underlying cause of pigmentation. It may be physiological or clinical manifestation of any underlying systemic disorder or due to medications or adverse habits. Here, we present a case report on lenticular pigmentation with emphasis on differential diagnosis.

KEYWORDS: Oral pigmentation, lenticular pigmentation, Addison's disease, Peutz-Jegher syndrome, Laugier- Hunziker syndrome

INTRODUCTION:

The normal oral mucous membrane is of varying shades of red. When the pigmentation area is noticed by the patient or the clinician, there is an element of increased concern. [1] Usually, an in-depth examination is required for focal lesions to exclude melanoma and a comprehensive examination for diffuse lesions. Some lesions show signs of diseases with systemic implications like adrenal insufficiency. [2] Any pigmented lesion on the oral mucosa will be considered as melanoma until evidence to the opposing. Due to possibility of similarities, histological examinations are necessary for any pigmented oral mucosa lesion to confirm or disprove the clinically suspected diagnostic hypothesis. [3]

CASE REPORT:

A 23-year-old male patient came to the Department of Oral Medicine and Radiology with a chief complaint of broken tooth in upper front teeth region of the jaw for past 10 years with a history of trauma. The patient's medical history was not significant. Personal history reveals usage of smokeless tobacco in labial mucosa of upper and lower lip for the past years, with a frequency of 3 times per day. General, systemic, and extra-oral examinations were not significant. On intraoral hard tissue examination, dental caries in 17, 27, 37, 47 and 48; fracture of enamel in 21 was noted. Soft tissue examination reveals multiple, discrete, lenticular, blackish-pigmented macules in upper labial mucosa (fig 1) and white non-scrapable lesion with wrinkled surfaces in the upper labial mucosa and related labial vestibule. Diffuse blackish pigmentation is seen in the left lower labial mucosa (fig 2). Based on the chief complaint, history, and clinical examination it was provisionally diagnosed as Ellis class 1 fracture in 21 and other diagnosis of Idiopathic pigmentation in upper and lower labial mucosa with a differential diagnosis of Addison's disease, Peutz-Jeghers syndrome, Laugier-Hunziker syndrome.

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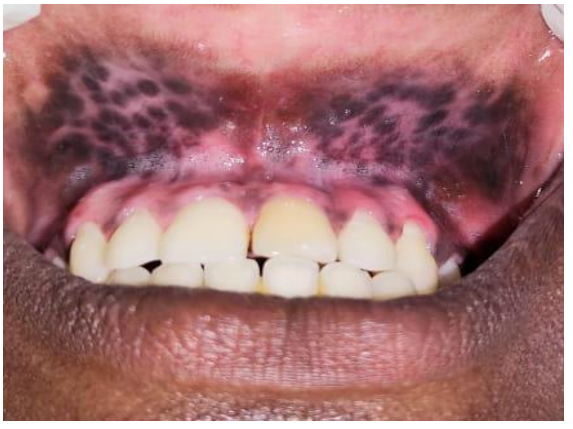

Fig 1: Multiple, discrete, lenticular blackish pigmented macules in the upper labial

DISCUSSION:

The differential diagnosis of oral pigmentations includes Laugier-Hunziker syndrome, Peutz- Jeghers syndrome, Addison's disease, and Pigmentation due to smoking and medication. PEUTZ-JEGHERS Syndrome (PJS) shows mucocutaneous pigmentation macules around the cheek and mouth, multiple hamartomatous polyps in the gastrointestinal tract, and family history (inherited autosomal dominant disease). Incomplete PJS is a condition with either mucocutaneous pigmentation or gastrointestinal polyps, which usually presents only typical pigmentation or enterorrhagia or symptoms of intussusception. The relation between PJS and STK11 gene has been confirmed by researchers. Jenne and Hemminki colonized a PJS-related gene, in 1998, and named it STK11. In Hungary, Papp J found about 21 cases from 13 families with the STK11 mutation. The patient did not have any family history. [4] ADDISON'S DISEASE is an autoimmune disease. This shows a specific sign of hyperpigmentation of It occurs due to inadequate production of cortisol and aldosterone as a result of destruction of the adrenal cortex skin and mucosal surfaces.

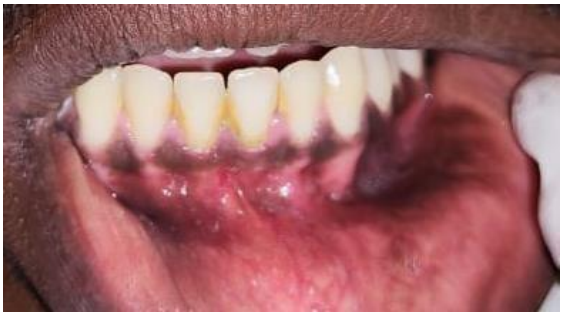

Fig 2: Diffuse blackish pigmentation in the lower labial mucosal region and gingiva

It affects all age groups and has no gender predilection. The pigmentation macules can be found diffusely on the gingiva, buccal mucosa, hard palate, and tongue. The macules tend to be blue- black or brown and can be spotty or streaked in the configurations. [5, 6] Recognizing the early signs of Addison's disease is critical because the condition can be fatal if not treated. The diagnosis involves exogenous ACTH stimulation testing with subsequent measurement of plasma ACTH and serum cortisol levels. Treatment involves steroid replacement therapy that usually resolves the hyperpigmentation. [7] There is no history of increased levels of ACTH. LAUGIER-HUNZIKER (LHS)syndrome is an idiopathic macular hyperpigmentation of skin. It is characterized by brownish-black spots on oral mucosa, including lips associated with longitudinal melanonychia of nails. [8] Oral hyperpigmentation may be the only presenting sign or may co-exist with skin and nail pigmentation. The most commonly involved site is the buccal mucosa and lower lip. They occur in a form that measures about 1.5 mm in size as smooth-surfaced brown, black, or slate-colour macules. Pseudo-Hutchinson sign when hyperpigmentation of the nail bed and matrix reflects through the transparent nail folds simulating Hutchinson's sign, a marker of sub-ungual melanoma, has also been reported in a few cases of LHS. [9] This condition is excluded as it did not show nail and skin pigmentation.

CONCLUSION:

Dentists should be aware of the various lesions to aid in the proper treatment plan. Although clinical suspicion is strong, the only way for a final diagnosis of a pigmented oral lesion is through a comprehensive intraoral examination during any consultation with the dental surgeon, complemented by a full dermatological examination as and when needed and to decrease the rate of mortality, and morbidity prompt treatment is required.

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