Rare case of pigmentation of the oral cavity: A case report

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ABSTRACT

Pigmentation of the tongue can occur due to various physiological or pathological events with no significant difference between males and females and age groups. Change in the color of the tongue can be due to localized lesions or systemic disorders. Oral pigmentation has been associated with a variety of endogenous and exogenous etiologic factors. This case report was about a 40-year-old male patient who presented with fever and pigmentation of tongue and lips. In view of his pigmentation and sudden deterioration in condition, a diagnosis of Addison's crisis was considered. A computerized tomography scan of his abdomen showed a hypodense non-enhancing lesion in the right adrenal gland, suggestive of tuberculosis. He was started on steroids and antitubercular regimen of rifampicin, isoniazid, ethambutol, and pyrazinamide. After 1 month of treatment, the pigmentation of the tongue and the lips started decreasing.

Key words: *Oral mucosa, Pigmented tongue, Tuberculosis*

Pigmentation is a process of deposition of pigments in various tissues. Human oral pigmentation may be physiological or pathological. The term "pigmentations of the oral mucosa" may be applied to a wide range of entities caused by the accumulation of one or more pigments and featuring a change in color of the tissues [1,2]. The intensity and distribution of pigmentation of the oral mucosa are variable not only between races but also between different individuals of the same race and within different areas of the mouth. Most of the pigmentations are caused by five primary pigments. These include melanin, melanoid, oxyhemoglobin, reduced hemoglobin, and carotene. Others are caused by bilirubin and iron [3-5].

Melanin is produced by melanocytes in the basal layer of the epithelium and is transferred to the adjacent keratinocytes through membrane-bound organelles called melanosomes. Melanin is also synthesized by nevus cells, which are derived from the neural crest and is found in the skin and mucosa. Pigmented lesions caused by increased melanin deposition may be brown, blue, gray, or black, depending on the amount and location of melanin in the tissues. Pigmentation can be produced by various drugs such as hormones, oral contraceptives, chemotherapeutic agents such as cyclophosphamide, busulfan, bleomycin, and fluorouracil, tranquilizers, antimalarials such as clofazimine, chloroquine, and amodiaquine, antimicrobial agents such as minocycline, antiretroviral agents such as zidovudine, and antifungals such as ketoconazole. Palate and gingiva are the most common sites affected.

We present a case of adrenal gland tuberculosis presenting as pigmentation of the tongue and lips.

CASE REPORT

A 40-year-old man presented to our hospital with a complaint of fever for 20 days. He felt chills at times and a sense of increasing weakness that he noticed for the past 10 days. He had lost his appetite and was at times nauseated. There was no history of cough, dysuria, and diarrhea abdomen/chest pain, headache, and joint pains. He had no similar history in the past. The patient was not on any medication for present or past illness.

On general examination, he was conscious oriented and alert. He had a temperature of 100.8 degrees Fahrenheit in the morning with a heart rate of 110/min and a blood pressure (BP) of 90/60 mmHg. There was mild pallor, no cyanosis clubbing, or icterus. The neck was soft, no glands were palpable superficially, and there was no hepatosplenomegaly. The chest was clear and there was no murmur. On local examination of the oral cavity, pigmentation was seen on the dorsal aspect of the tongue, predominantly occupying the anterior one-third of the tongue and also on upper and lower lips (Fig. 1). The pigmentation was dark gray to black in color and moderate in intensity. No alteration in the pigmentation with intake of food and also no history of drug use.

He underwent workup for pyrexia of unknown origin. The complete blood counts and renal and liver functions were normal. No parasites were found and the blood and urine cultures were negative. The chest radiograph was normal and the ultrasound of the abdomen showed mild hepatosplenomegaly.

On the 3rd day of admission, he became anxious and sweaty. His pulse increased without fever and his systolic BP started

falling. He was shifted to the intensive care unit. In view of his pigmentation and sudden deterioration in condition, a diagnosis of Addison's crisis was considered. His serum cortisol was sent at 8 am and started on steroids. His condition improved dramatically. A computerized tomography (CT) scan of his abdomen was done. It revealed bilateral bulky adrenals with standing of fat. The right adrenal gland also showed a hypodense non-enhancing lesion, suggestive of tuberculosis (Fig. 2). His serum cortisol was <0.2 (reference range 5–25 mcg/dL). He was started on steroids and antitubercular regimen of rifampicin, isoniazid, ethambutol, and pyrazinamide.

The patient was called for follow-up every month and examined for the various symptoms and signs including oral pigmentation. The pigmentation of the tongue and the lips started decreasing after 1 month of antitubercular drugs and a repeat CT of the abdomen was done at the 6th month of treatment which showed complete resolution of the lesion in the adrenals (Fig. 3).

DISCUSSION

Pigmentation of the tongue and lips may be physiological or pathological and exogenous or endogenous. Endogenous pigmentations of the oral mucosa are produced by the body's own metabolism defects. These may include melanin, hemoglobin, and hemosiderin. The most important of them is melanin, which is synthesized by melanocytes in the basal epithelial layer and then transferred to keratinocytes. There is more occurrence of melanin pigmentation in the oral cavity of darker-skinned individuals than light-skinned individuals. The coloration is produced by melanocytes that contain melanin in the basal cell layer of epithelium. It produces melanin in membrane-bound organelles called as melanosomes [6]. Exogenous pigmentation is due to exposure to various heavy metals such as mercury, lead, graphite, bismuth, and silver amalgam [7].

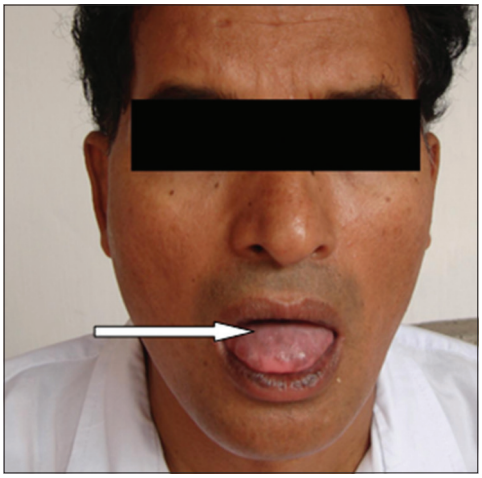

Figure 1: Patient with pigmented tongue

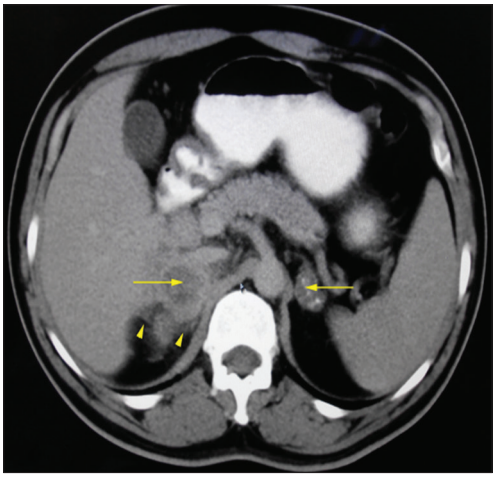

Figure 2: Computerized tomography scan showing adrenals with tuberculous lesion

Addison's disease, also known as primary adrenal insufficiency and hypocortisolism, is a long-term endocrine disorder, in which the adrenal glands do not produce enough cortisol [8]. Symptoms generally come on slowly and may include abdominal pain, weakness, and weight loss. Darkening of the skin or mucosal membrane in certain areas may also occur. Autoimmune adrenalitis is the most common cause of Addison's disease.

Autoimmune destruction of the adrenal cortex is caused by an immune reaction against the enzyme 21-hydroxylase [9]. This may be isolated or in the context of the autoimmune polyendocrine syndrome Type 1 or 2, in which other hormone-producing organs, such as the thyroid and pancreas, may also be affected [10]. Adrenal destruction is also a feature of adrenoleukodystrophy when the adrenal glands are involved in metastasis (especially lung), hemorrhage, and in some specific infections such as tuberculosis, histoplasmosis, coccidioidomycosis, or amyloid deposition [11].

In this case, the patient presented with the pigmentation of the tongue and the lips. His serum cortisol was sent at 8 am and started on steroids. His condition improved dramatically. A CT scan of his abdomen was done. It revealed bilateral bulky adrenals with standing of fat. The right adrenal gland also showed a hypodense non-enhancing lesion, suggestive of tuberculosis. The patient's condition improved further and the pigmentation of the tongue disappeared after antituberculous treatment. A similar case of pigmentation with tongue ulcer was reported as tuberculosis by Kim *et al.* [12].

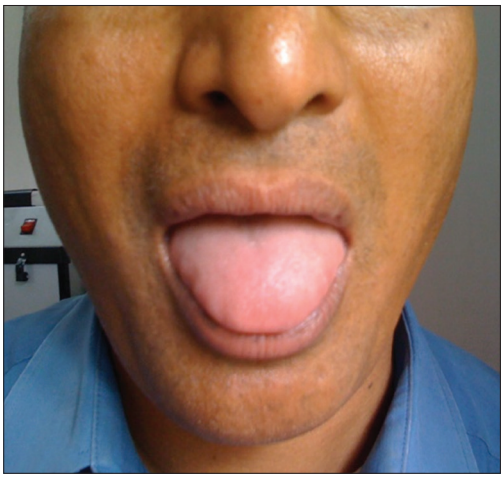

Figure 3: Patient after the treatment of pigmented tongue

Differential diagnoses include the use of medications such as quinine drugs, smoker's melanosis, heavy metal poisoning with lead, arsenic, gold, bismuth, Peutz–Jeghers syndrome, candidiasis, thyrotoxicosis, lamb and leopard syndrome, neurofibromatosis, polyostotic fibrous dysplasia, the McCune-Albright's syndrome, the Jaffe-Lichtenstein syndrome, and Addison's disease.

CONCLUSION

The difference in color of the oral tissue should attract the clinician's attention as these could be due to the pathological process. The diagnosis of pigmented lesions of the oral cavity is a very challenging task for the clinician. Higher investigation modalities should be used for early diagnosis and treatment. In this case, a CT scan of the abdomen was helpful in diagnosis the disease entity and the cause for pigmented tongue and lips.

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