Approach to Tongue Hyperpigmentation in General Practice: Pigmented Fungiform Papillae and Complexion-Associated Melanosis in a Young Pakistani Female

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Abstract

Hyperpigmentation in exposed areas like the tongue and conjunctiva can pose diagnostic challenges in general practice, ranging from serious conditions to benign, aesthetic concerns. This case report examines a 21-year-old female with concurrent benign hyperpigmentation of the tongue and conjunctiva. A thorough evaluation ruled out extrinsic causes and systemic illnesses, leading to the diagnosis of Pigmented Fungiform Papillae (PFP) on the tongue and Complexion Associated Melanosis (CAM) in the conjunctiva. Due to the absence of concerning features, biopsy was deferred in favor of routine surveillance. An inquiry into the potential association between hyperpigmentation and conditions such as anemia and vitamin D deficiency revealed no definitive connection, as correcting these deficiencies did not resolve the hyperpigmentation. This case underscores the importance of recognizing benign hyperpigmentation to avoid unnecessary procedures and emphasizes a cost-effective, patient-centered approach.

Keywords: *Pigmented fungiform papillae, tongue hyperpigmentation, complexion-associated conjunctival melanosis, iron deficiency anemia, tongue melanosis, black tongue.*

INTRODUCTION

Hyperpigmentation disorders primarily result from hypermelanosis, characterized by increased melanin biosynthesis and deposition in the epidermis and dermis. In the tongue, hyperpigmentation may stem from both acquired and genetic factors, or it may indicate more serious underlying conditions such as Addison's disease, melanoma, or diffuse melanosis cutis [1, 2]. Among benign differentials, Pigmented Fungiform Papillae (PFP) is a rare form of tongue hyperpigmentation [3-5], while Complexion Associated Melanosis (CAM) affects the ocular conjunctiva [6]. This report presents a case of a young woman with tongue and conjunctival hyperpigmentation, accompanied by iron and vitamin D deficiencies. It suggests an approach for managing such patients in general practice based on recent literature.

CASE PRESENTATION

A 21-year-old woman presented with hyperpigmented patches on her tongue and darkening of the bulbar conjunctiva bilaterally, which had progressed over 2-3 years. She reported no pain, itching, change in taste, halitosis, or vision changes. Recent symptoms included shortness of breath on exertion and significant hair loss. There were no reports of weight loss, chest pain, palpitations, perspiration, oral ulcers, photosensitivity, joint pains, or menstrual changes. Menstrual cycles were regular. Her past medical history included a previous episode of anemia, with no further details recalled. The patient was not on any medication and denied recent dietary changes. Family history included hypertension and diabetes but no hyperpigmentation disorders. On examination, she appeared pale, with a blood pressure of 133/87 mmHg and a pulse rate of 98 bpm. The oral examination revealed well-defined, asymmetric black-pigmented patches on the dorsum of the tongue (**Fig. 1**), sparing the rest of the oral mucosa. Ocular examination showed bilateral brown flat lesions with ill-defined margins on the bulbar conjunctiva (**Fig. 2**). Examination of other organ systems was unremarkable. Based on her history and examination findings, the following tests were done (**Table 1**).

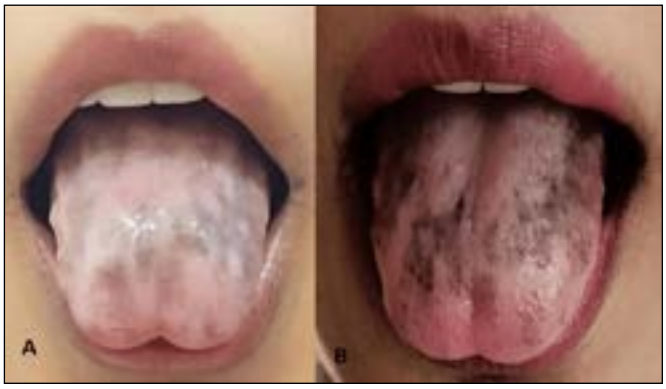

Fig. (1): Tounge Hyperpigmentation: **A** First Visit **B** Follow-up at four months.

Based on the history, examination, and investigations, our differentials for tongue hyperpigmentation narrowed down to localized hyperpigmentation, which was deemed acquired and benign due to its stable course over observed time, leading to a watchful waiting

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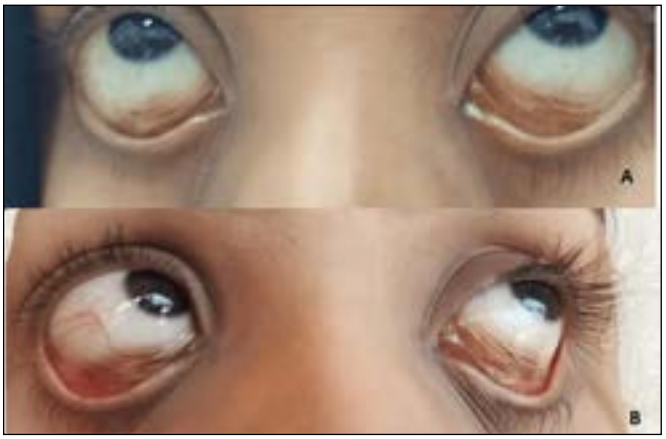

Fig. (2): Conjunctival Hyperpigmentation: **A** First Visit **B** Follow-up at four months.

Table 1: Investigations.

Investigation	Patient Values	Reference Range
Hb	7 g/dL	12 to 15 g/dL
MCV	50 fL	80–100 fl
WBC	6.5 x 10^3/uL	4 to 10 x 10^3/uL
Platelets	511 x 10^3/uL	150-400 x 10^3/uL
Cr	0.57 mg/dL	0.6 to 1.1 mg/dL
Vitamin D	13.62 ng/mL	20 to 40 ng/mL
Na	141 mmol/L	136 to 146 mmol/L
K	3.8 mmol/L	3.5 to 5.1 mmol/L
Cl	106 mmol/L	98 to 107 mmol/L
HCO3	23.7 mmol/L	22 to 29 mmol/L
Serum Iron	9 µg/dL	40 to 160 µg/dL
TIBC	423 µg/dL	250-400 µg/dL
TSH	4.250 µIU/mL	0.4 to 4.2 mIU/L
Free T4	1.06 ng/dL	0.8 to 1.8 ng/dL
Serum Cortisol (8 am)	20.36 µg/dL	5 to 25 mcg/dL
ACTH	29 pg/m	10 and 60 pg/mL

Hemoglobin (Hb), Mean Corpuscular Volume (MCV), White Blood Cell count (WBC), Platelet count (Plt), Creatinine (Cr), Vitamin D, Sodium (Na), Potassium (K), Chloride (Cl), Bicarbonate (HCO_3), Serum Iron, Total Iron- Binding Capacity (TIBC), Thyroid-Stimulating Hormone (TSH), Free Thyroxine (Free T_4), Serum Cortisol (8 am), and Adrenocorticotropic Hormone (ACTH).

approach. We diagnosed the patient with microcytic anemia, primarily due to iron deficiency, along with concurrent vitamin D deficiency. Treatment included an iron-rich diet, iron supplements, and vitamin D replacement. We aimed to observe whether anemia and vitamin D deficiency were linked to hyperpigmentation. However, despite improvement in anemia and vitamin D levels, the hyperpigmentation of the tongue and conjunctiva persisted at the one-year follow-up. The hyperpigmentation resembled Pigmented Fungiform Papillae (PFP) on the tongue and Complexion Associated Melanosis (CAM) on the conjunctiva (**Fig. 1** and **2**).

DISCUSSION

Hyperpigmented lesions are typically diagnosed based on clinical factors such as size, shape, color, and duration, with biopsy employed when necessary, in correlation with the patient's medical history [1, 2]. Tongue hyperpigmentation may be either inherited, as

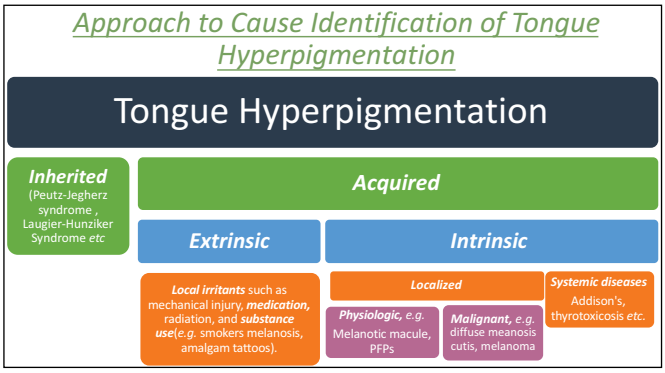

Fig. (3): Alogorithmic approach to tongue Hyperpigmentation.

seen in Peutz-Jeghers Syndrome and familial Laugier-Hunziker syndrome [7-9], or acquired due to internal or external factors like medication, radiation, injury, or substance use, which can lead to potentially reversible hyperpigmentation [10-14].

Localized hyperpigmentations, such as macula and pigmented fungiform papillae, are often physiological and linked to increased melanocyte activity, particularly in individuals of African, Asian, or Mediterranean descent. Systemic conditions, such as elevated MSH and ACTH levels in Addison's disease, can also cause tongue hyperpigmentation. While generally benign [2, 15], hyperpigmentation can sometimes be indicative of malignant disorders such as melanoacanthoma and melanoma [1, 2].

When systemic illnesses and external factors are ruled out, a biopsy can be considered warranted to rule out melanoma, especially if there are concerning signs such as rapid changes in color, size, form, or poorly defined borders [2].

In the present case, a comprehensive medical history and examination were performed to ensure a thorough understanding of the patient's condition. The patient denied any medication, substance use, or other exposures, and the family history was non-contributory. The absence of characteristic darkening in other body parts, such as acanthosis nigricans in Addison's disease or sparing of the lips and remaining oral mucosa in Peutz-Jeghers syndrome, initially ruled out inherited and extrinsic causes [2, 7] (**Fig. 3**).

Relevant diagnostic tests, including serum cortisol, ACTH, TSH, and electrolytes, were conducted to evaluate systemic causes such as Addison's disease and thyroid disorders. As these tests did not indicate any abnormality, further diagnostic steps, such as biopsy, were considered. However, the literature suggests that most hyperpigmentation disorders are benign, more common in dark-skinned populations, including Asians, and can be diagnosed clinically [1, 15]. Since the patient's hyperpigmentation lacked concerning features, watchful waiting was adopted as a management strategy in shared decision-making with the patient.

Over time, the patient did not report any rapid changes in the color, size, or shape of the hyperpigmented lesions, which were deemed acquired and benign. Given the distribution of lesions along the anterior and lateral borders of the tongue, accompanied by prominent pointed papules, a clinical diagnosis of Pigmented Fungiform Papillae (PFP) was established [3-5]. Similarly, Complexion-associated melanosis (CAM), a benign epithelial conjunctival melanosis with a negligible risk of malignancy, was considered the likely cause of hyperpigmentation in the bulbar conjunctiva due to its characteristic appearance, bilateral distribution, and benign course [6]. Hypermelanosis is the suggested histological feature of both PFP and CAM. Based on the observed course of these lesions and their reported negligible risk of malignancy, a biopsy was deferred, and routine surveillance was recommended for future follow-ups [3-6].

The authors explored potential connections between conjunctival melanosis (CAM), tongue hyperpigmentation (including PFP), anemia, and vitamin D deficiency. Three of these four conditions (excluding vitamin D deficiency) were observed in a pediatric patient in India [16]. Anemia was noted in another recent case of tongue hyperpigmentation in a Saudi woman [5]. However, anemia is a common condition, and it is noteworthy that addressing these deficiencies in our patient did not lead to a reversal of these hyperpigmentations during the observation period of one year, further defying any definitive connection between these conditions. Similar to our case, a recent review on PFP reported that most cases occur in females, primarily under 40 years of age [3-4].

Although PFP and CAM are benign physiologic hyperpigmentation conditions with no significant adverse effects, the exact cause of simultaneous physiologic hyperpigmentation in multiple areas remains unknown. Clinicians should familiarize themselves with these conditions to promote a cost-effective approach, avoid unnecessary invasive procedures, and minimize negative impacts on patients' perceptions.

CONCLUSION

In conclusion, a diagnosis of physiological hyperpigmentation, such as PFP and CAM, can be considered on clinical grounds with a watchful waiting approach in cases with hyperpigmentation of the tongue and bulbar conjunctiva, provided that other extrinsic and systemic causes are ruled out based on a comprehensive evaluation. As per our case, improvement in anemia and vitamin D deficiency were found to have no effect on the resolution of hyperpigmentations, defying any connection between them.

CONSENT FOR PUBLICATION

Informed consent was taken from the patient.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

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Declared none.

AUTHORS' CONTRIBUTION

FK: Substantial contribution to the conception of work, collection of data from patient, literature review, drafting manuscript and agreement to be accountable for all aspects of the work, and approval of the final version.

WS: Substantial contribution to the conception of work, literature research, data collection, drafting the manuscript, revising it critically for important intellectual content, and approval of the final version to be published, agreement to be accountable for all aspects of work.

AB: Interpretation of data, revising it critically for important intellectual content, final approval, and agreement to be accountable for all aspects of work.

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