Case Report

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Pigmented fungiform papillae in a middle-aged Saudi female

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Abstract:

Pigmented fungiform papillae are a rare benign condition. It is commonly considered a diagnosis dilemma as it could be a clinical sign of a serious condition. Our patient was a middle-aged Saudi female presenting with hyperpigmentation along the lateral and anterior aspects of her tongue and was not associated with other cutaneous or mucocutaneous manifestation. Her medical history was significant for iron deficiency anemia, hepatic hemangioma, cervical and lumbar intervertebral disc herniation, and gastroesophageal reflux disease. Upon inspection, she had a longitudinal hyperpigmented tan-brown patch on the lateral aspects of her tongue and pinpoint discoloration evenly distributed on the papillae of the anterior tongue. The examination by mucoscopy revealed a cobblestone appearance along the mentioned distribution. The patient refused to have the biopsy done, but she was reassured about her condition and instructed to visit the clinic if she has any further issues.

Keywords:

Fungiform, mucous membrane, papillae, pigmented

Introduction

The tongue is covered by the four forms of papillae: Fungiform, filiform, circumvallated, and foliate. The fungiform papillae are located predominantly on the ventral and lateral sides of the tongue. Oral mucosa predominant mucosal pigment is due to melanin, melanoid, oxyhemoglobin, hemosiderin, bilirubin, iron, and carotene.^[1]

Pigmented fungiform papillae (PFP) are a rare benign condition that presents as painless hyperpigmentation. The overall prevalence is 1%–3% and is higher among African Americans (33%). The clinical signs of PFP are usually misinterpreted as serious pathological conditions which result in an unnecessary workup.^[2]

Case Report

A 44-year-old married Saudi female, known case of gastroesophageal reflux disease

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and chronic iron-deficiency anemia, skin type IV, presented with tongue discoloration of 5 months duration located on the lateral and anterior aspects of her tongue. She had reported a numb sensation without any sensory or motor function loss. There was no pain, tingling, or burning sensation. The patient did not complain of any other skin discoloration. She did not use any drugs and did not smoke, chew tobacco, or drink alcohol. Her dental history was insignificant. During that visit, she reported multiple symptoms over the last year for which we consulted the internal medicine team. There were the episodes of diffused abdominal pain associated with heartburn, dyspepsia, cough, and a change in her voice. She complained of unexplained on/ off night sweats, heat intolerance, tremors, and palpitations. She had also had irregular menses and decreased period flow for 5 months mostly for being perimenopausal.

On general examination, the patient was pale and obese with a body mass index of 38. She had good oral hygiene with no gum

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discoloration, but she had a longitudinal hyperpigmented tan-brown patch on the lateral aspects of her tongue and pinpoint discoloration evenly distributed on the papillae of the anterior tongue, as presented in Figure 1a-c. No other mucocutaneous lesions were observed and the rest of the clinical examination was unremarkable.

One year prior, she had bilateral paresthesia in her legs. Upon further investigation, magnetic resonance imaging showed cervical prolapse in C1 through C6 and a lumbar prolapse in L5/S1. She was managed with tramadol and physiotherapy. About 3 years before an abdominal US revealed hepatic hemangioma. Finally, her screening mammography showed benign changes.

Laboratory work revealed iron-deficiency anemia and a very high ferritin of 931.21 ng/mL that spiked from 5 ng/mL in 1 month with no obvious clinical cause. Antithyroglobulin was 19.91; however, other thyroid parameters were within the normal range. Other immunoassays and immunology (anti-gliadin AB, anti-endomysial AB, etc.,) were negative. Her Vitamin D level was low (|17.8 ng/mL). Hepatitis B and C panels were negative.

Upon further follow-up, the mucoscopy showed a cobblestone appearance, as shown in Figure 2. The patient refused to have the biopsy done, but she was reassured about her condition and instructed to visit the clinic if she has any further issues.

Discussion

The majority of PFP cases reported were by females, although it is prevalent more in dark-skinned individuals, the reported cases were mostly Asian. In most cases, the pigmented papillae were present in anterior and lateral.^[1] There have also been reports of pigmentation in the proximal nail folds and gums.^[3]

PFP is divided into three different categories: The patient in this case was categorized as type 1, which was distinguished by clearly defined hyperpigmented patches

including the fungiform papillae on the anterolateral side and tip of the tongue. Type 2 is categorized by a few sparse fungiform papillae that are hyperpigmented in the dorsal surface of the tongue. Type 3 is characterized by a diffused darkening of all fungiform papillae on the tongue's dorsum.^[1]

The causes of mucosal pigmentation are categorized as pathological or physiological. Exogenous pathological causes include drugs, smoking or chewing tobacco, and amalgam tattoos. Inflammatory conditions such as lichen planus, Peutz-Jeghers disorder, or endocrinological conditions such as Addison's disease, diabetes mellitus, pregnancy, and hyperthyroidism are the examples of endogenous causes.[4,5] Although many patients do not have other complaints, some recorded cases have particular correlations, including linear circumflex ichthyosis, lichen planus, Hori's nevus, melisma, hemochromatosis, scleroderma, pernicious anemia, and iron-deficiency anemia as obtained in our patient.^[1,5] There are correlations linking pigmented papillae fungiform to high levels of estrogen as menstrual irregularities and obesity, which is a possible etiology in this case.^[1,6] PFPs are often described as an oral manifestation of Laugier-Hunziker syndrome, a rare disorder that causes mucocutaneous pigmentations. The differential diagnosis of PFP includes a black hairy tongue, median rhomboid glossitis, benign migratory glossitis, macula melanotic, physiological hyperpigmentation, lingual varicosity, and leukoplakia.^[1]

Although most cases of PFP are diagnosed clinically, mucoscopy helps in describing PFP as having a cobblestone appearance in 47.37% and a rose petal appearance in 52.63%.^[1,4,7] Our case showed a cobblestone appearance, which is light to dark brown pigmentation on papillae, round or polygonal in shape, and circumscribed. Rose petal appearance is described as pigmented borders, ovoid hyperpigmented projections with dichotomized vessels originating at the base.



Figure 1: (a) Pigmented fungiform papillae on the tongue at the anterior side. (b) Pigmented fungiform papillae on the tongue at the left lateral side. (c) Pigmented fungiform papillae on the tongue at the right lateral side

Immunohistochemical stains such as melan-A and S-100 protein are useful in ruling out more serious causes such as melanoma.^[8] Microscopic examination of PFP



Figure 2: Cobblestone appearance on mucoscopy

shows deposition of melanin, hyperpigmentation of basal keratinocytes, and melanophages in the lamina propria. Lymphocytes are also present in the connective tissue stroma.^[1,4]

Two cases of PFP reported with lingual fimbriae can sometimes be confused with squamous papilloma or oral warts.^[9] There is no reported cure, although one case reported an improvement after the treatment of iron-deficiency anemia.^[5]

Conclusion

Tongue lesions are seen frequently in primary healthcare. Benign conditions such as PFP should be kept in mind as well as other pathological causes to avoid an overload of histological examinations and other unnecessary workup.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given

her consent for her images and other clinical information to be reported in the journal. The patient understands that name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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