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# Hyperpigmented fungiform papillae of the tongue: A report of 2 pediatric cases

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## Abstract

**Introduction:** Pigmented Fungiform Papillae of the Tongue (PFPT) also known as hyperpigmented fungiform papillae of the tongue is a benign, asymptomatic, and non-progressive condition predominantly affecting individuals with dark skin types. It typically appears between the second and third decades of life, although isolated cases have been described in pediatric patients.

**Case description:** We present 2 cases of male patients aged 10 and 9 years, respectively, who were incidentally found to have lingual fungiform papillary hyperpigmentation during oral cavity examination. One of them also had associated pigmentation of the gums. Neither presented with associated systemic symptoms, leading to a diagnosis of lingual fungiform papillary hyperpigmentation, with an explanation provided regarding the benign nature of the finding.

**Discussion:** The etiopathogenesis of PFPT is unclear, although it is postulated to be related to female sex hormones given its higher prevalence in women. Differential diagnosis should include Addison's disease, melanocytic nevus, amalgam tattoo, black hairy tongue, Laugier-Hunziker syndrome and Peutz-Jeghers syndrome. PFPT does not require complementary tests, follow-up, or treatment. In cases where patients have significant aesthetic concerns, laser treatments may be offered for hyperpigmentation removal.

**Conclusions:** Due to increasing migration, we are likely to see more cases of PFPT in Europe. It is important to identify this incidental finding and recognize it as a variant of normality to avoid unnecessary additional testing.

***Keywords:*** Fungiform Papillae; Pigmented; Tongue; Pediatric.

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## Introduction

Fungiform papillae of the tongue are localized projections on the dorsal surface of the tongue, predominantly found on the dorsal and lateral regions, and they are part of the taste papillae. PFPT also known as hyperpigmented fungiform papillae of the tongue is a benign, asymptomatic, and non-progressive condition that predominantly affects black individuals. Although there are some reported cases in Asian and Indian populations, it is considered rare in Eastern races and exceptional in individuals of white race [1]. It typically appears around the second or third decades of life, although isolated pediatric cases have been described. PFPT is a rare idiopathic condition that is sometimes associated with hyperpigmentation of the gums and nail folds [2]. It does not require treatment, but understanding its existence is important as it allows for clinical diagnosis, thereby avoiding unnecessary additional testing [3].

## Case descriptions

### Case 1

A 10-year-old male patient of Latin descent with Fitzpatrick skin type IV presented to the clinic with symptoms of odynophagia. Upon physical examination, mild pharyngeal erythema was noted along with the presence of multiple hyperpigmented macules, measuring 0.5-1 mm in diameter, distributed on the distal third and lateral regions of the dorsal surface of the tongue (Figure 1); the remainder of the physical examination was normal with no hyperpigmentation observed in other locations.

The patient reported not being aware of when the hyperpigmentation had appeared, and indicated that his immediate family members did not have similar findings. Given the distribution of the hyperpigmentation and absence of other systemic symptoms, a diagnosis of hyperpigmented fungiform papillae of the tongue was made and the benign nature of the finding was explained.

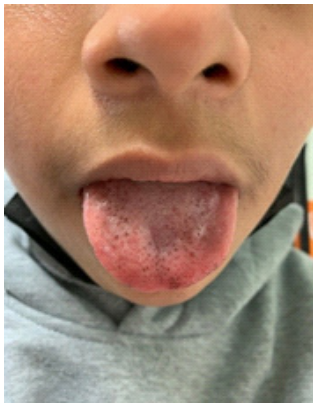

**Figure 1:** Clinical photograph of tipe I PFPT according to Holzwanger´s classification. The patient has well-defined macules on the anterior and tip of the tongue surface.

### Case 2

A 9-year-old male patient of African descent with Fitzpatrick skin type VI presented to the clinic with a complaint of molar pain. Upon physical examination, the only notable findings were the presence of hyperpigmented macules of approximately 0.5 mm in diameter on the distal third of the dorsal surface of the tongue (Figure 2), along with hyperpigmentation of the gums (Figure 3); the rest of the physical examination was normal.

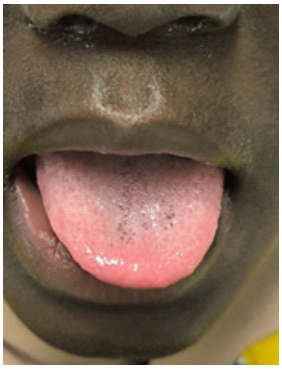

**Figure 2:** Clinical photograph of tipe II PFPT according to Holzwanger´s classification. The patient has several fungiform papillae pigmented on the dorsal surface of the tongue.

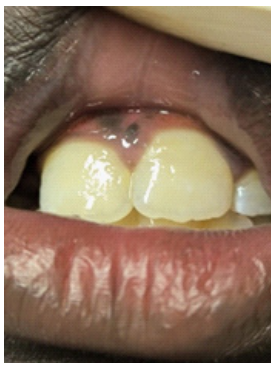

**Figure 3:** Clinical photograph showing hyperpigmentation at the gum level.

A diagnosis of hyperpigmented fungiform papillae of the tongue was made, and the family was informed about the benign nature of this condition. No treatment was recommended.

## Discussion

Pigmented Fungiform Papillae of the Tongue was first described in 1905, initially thought to be related to hookworm infection. Subsequently, other authors have observed it in association with other conditions such as circinate erythema, lichen planus, hemochromatosis, scleroderma, pernicious anemia, and iron deficiency anemia [1]. However, these associations are proposed based on isolated cases where it appears to be an incidental finding, as Kaplan reported an incidence of 6% in Black men and 8% in Black South African women, and Holzwanger et al. [4] reported that 30% of Black women and 25% of Black men in the US had PFPT. Tan et al. [5] reported that 0.4% of all outpatient patients in China had PFPT.

The pathogenesis of this condition is unknown; however, it is postulated to be related to female sex hormones because most reported cases are in obese women and/or associated with early menarche [3]. In our experience, we have only observed this pigmentation alteration in males, so we cannot support or refute this hypothesis.

Histologically, in the pigmentation of fungiform papillae, numerous melanophages are observed in the chorion of the fungiform papillae, without the presence of inflammatory infiltrate.

The pigment located inside the melanophages shows positivity for melanin with Fontana-Masson staining and negativity for iron with Prussian blue staining. The acquired nature of the lesions and the presence of melanophages suggest a transient period of inflammation, but the lack of inflammatory infiltrate is one of the histological markers of the entity [1].

The age of onset of symptoms varies around the second or third decade of life, although isolated cases of onset in pediatric age have been described; in our case, both patients started experiencing symptoms in pediatric age. This condition persists over time but is not progressive [3].

Hyperpigmentation can appear in 3 patterns according to Holzwanger's classification [4]: I) Well-defined macules on the anterior, lateral, or tip of the tongue surface; II) Hyperpigmentation affecting several fungiform papillae on the dorsal surface of the tongue; III) Hyperpigmentation of all fungiform papillae on the dorsal surface of the tongue. In our case, the 10-year-old patient (Figure 1) would follow the distribution of pattern I, and the 9-year-old patient (Figure 2) would follow the distribution of pattern II according to Holzwanger´s classification.

The differential diagnosis should be made with Addison's disease, melanocytic nevus, amalgam tattoos, and hairy black tongue. Some genodermatoses such as Laugier-Hunziker syndrome and Peutz-Jeghers syndrome should also be considered in the differential diagnosis [3]. Once pathology is ruled out, it is important to explain to patients and families that this is a benign condition that does not require additional testing, follow-up, or treatment. In some cases where patients may have significant aesthetic concerns due to this condition, laser treatments can be offered for hyperpigmentation removal [6].

## Conclusions

Due to increasing migration, we will continue to see more cases of PFPT in Europe, making it important to recognize this incidental finding and understand that it represents a variant of normality. This awareness is crucial in order to avoid unnecessary additional testing.

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