

## Two Cases of Pigmented Fungiform Papillae of the Tongue

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**Pigmented fungiform papillae of the tongue (PFPT) is a rare benign pigmentary disorder of the tongue. In dark-skinned individuals, PFPT appears to be relatively common, whereas this entity is seldom reported in fair-skinned people and Asians. Although the relevance to systemic conditions has been suggested in several reported cases, the etiology and pathogenesis of this entity are not yet clearly defined.**

**We report two cases of PFPT in young Korean females. Clinically both patients presented with asymptomatic pigmentary lesions confined to fungiform papillae of the tongue, which had been detected several months ago. Histopathologic findings revealed increased melanophages in the subepidermal area within the fungiform papillae. (Ann Dermatol 14(4) 216~219, 2002).**

*Key Words* : Pigmented fungiform papillae of the tongue, Pigmentary disorder, Tongue, Asians

Pigmented fungiform papillae of the tongue (PFPT), which was first described by Leonard<sup>1</sup> in 1905 as a putative adverse effect of human hookworm disease, is characterized clinically by pigmentation confined to the fungiform papillae and histopathologically by the presence of numerous melanophages in the lamina propriae<sup>2</sup>. Although this condition is a rare pigmentary disorder in Asian people,<sup>3,4</sup> a few cases of Korean patients have recently been reported in dermatologic literatures<sup>5-7</sup>. We report herein two cases of PFPT in young Korean females.

### CASE REPORT

#### Case 1

A 25-year-old female presented with a hyperpigmented area on the dorsum of the tongue that had

been found accidentally several years ago. The lesion was totally asymptomatic, but had gradually spread. She was otherwise healthy and had no family history of a similar problem. There was no history of physical trauma, photosensitivity, specific medication, or any kinds of dental procedure. Physical examination revealed multiple dark brownish pigmentation confined to fungiform papillae on both anterolateral sides of the tongue (Fig. 1a). There was no evidence of oral mucous membrane hyperpigmentation elsewhere, nor was regional lymph node enlargement. The results of laboratory tests including complete blood cell count, blood chemistry, and urinalysis, were all within normal limits or negative. Histopathologic examination showed numerous melanophages in the upper lamina propriae of fungiform papillae (Fig. 1b). After a diagnosis of PFPT was made, no specific treatment was done.

#### Case 2

A 33-year-old woman presented with an asymptomatic hyperpigmented lesion on the tip and both anterolateral surfaces of the tongue, which had appeared several years ago. She denied noticeable medical problem or family history. Clinical manifestation was somewhat different from that of

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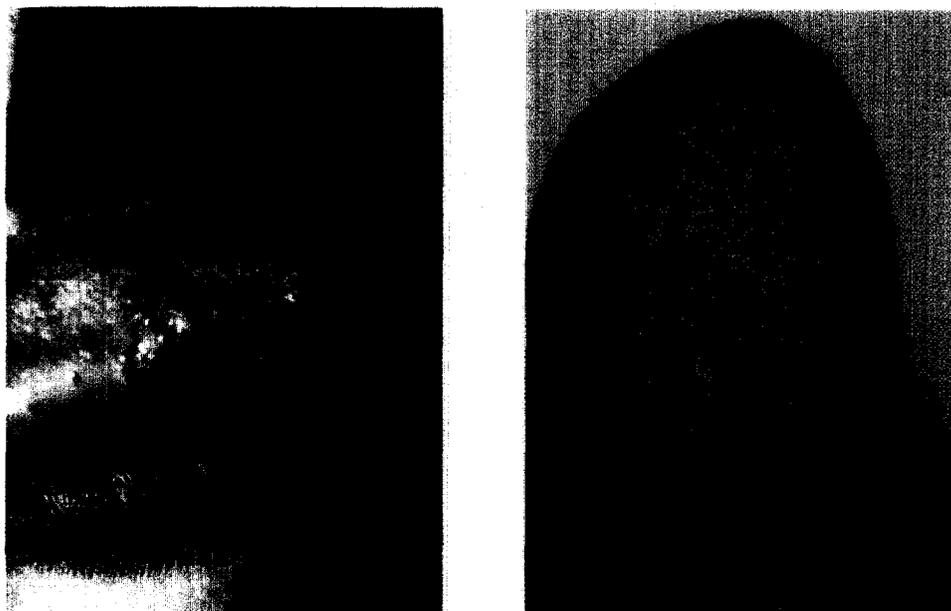


Fig. 1. Case 1. (a) Multiple dark brownish pigmentation confined to fungiform papillae on the anterolateral side of the tongue. (b) A biopsy specimen showing numerous melanophages in upper dermis of fungiform papilla (H&E,  $\times 100$ ).

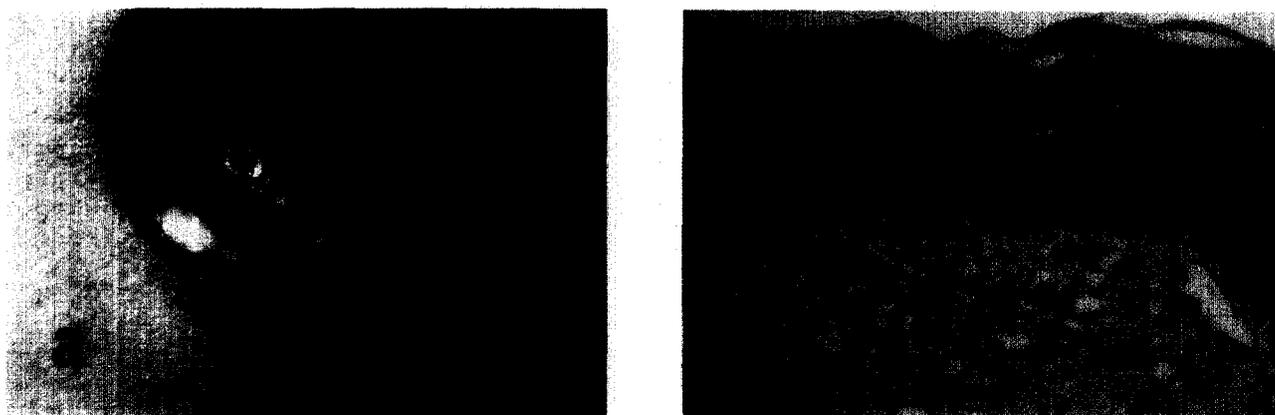


Fig. 1. Case 2. (a) Relatively ill-defined brownish macules with scattered darkly pigmented papillae were located on the anterolateral surface of the tongue. (b) High magnification of biopsy revealed upper dermal melanophages evidently (H&E,  $\times 400$ ).

case 1. Relatively ill-demarcated brownish macules were located on both anterolateral sides of the tongue (Fig. 2a). Darkly pigmented lingual papillae were scattered on the lingual tip and in the macular lesions of dorsal surface. Histopathologic findings were similar to those of case 1 and diagnosis was consistent with PFPT (Fig. 2b).

## DISCUSSION

PFPT (pigmented fungiform papillae of the tongue) is a benign pigmentary disorder, which occurs in persons of all races, but appears to be fairly common in black patients and comparatively rare among the yellow and the white races<sup>2,3</sup>. In

1973, Holzwanger et al<sup>2</sup>. performed a random survey of 300 individuals and noted that 30% of women and 25% of men had PFPT in 200 black people, but there was only one man with PFPT in 100 white people. He divided PFPT into three clinical types<sup>2</sup>. The first type was that of our second patient, i.e., a well-circumscribed hyperpigmented patch in which all the fungiform papillae were involved, located on the anterolateral aspect or tip of the tongue. The second type, which was similar to that of our first case, consisted of hyperpigmentation involving 3 to 7 fungiform papillae randomly scattered on the dorsal lingual surface. And the third type was hyperpigmentation of all fungiform papillae on the dorsum of the tongue.

Recently, cases of PFPT have been reported in Korea and Japan<sup>3-7</sup>. Hiramatsu et al<sup>4</sup>. reviewed 6 Japanese cases of PFPT to report that all 6 cases were female and the most common clinical pattern was the second type of PFPT. They also noticed that histologically all of their cases showed melanophages in the lamina propriae and half of them showed increased melanin pigmentation of basal cell layer and inflammatory cell infiltration in upper dermis<sup>4</sup>. On analyzing all Korean and Japanese cases, we observed that Asian patients showed absolute female predominance with the first type of PFPT as the most common clinical manifestation, while black people featured slight female predominance and the second type is more common than the first one in Holzwanger's study<sup>2</sup>.

PFPT may be easily diagnosed clinically, but most cases are thought to be unintentionally overlooked<sup>5</sup>. The differential diagnosis of oral hyperpigmentation is quite extensive. Isolated tongue hyperpigmentation is rare and has been associated with endogenous causes such as black hairy tongue, PFPT, melanoma, oral melanosis, physiologic melanin pigmentation, and as a normal variant. Exogenous causes include amalgam tattoo, chemotherapeutic agents, methyldopa, tricyclic antidepressants, and antibiotics such as minocycline<sup>8</sup>. Lingual hyperpigmentation can be associated with systemic conditions including Addison disease, Peutz-Jeghers syndrome, heavy metal poisoning, hemochromatosis, pernicious anemia, and scleroderma<sup>2,5,8,9</sup>. The main consideration in the differential diagnosis is black hairy tongue<sup>9</sup>, which has been attributed to the growth of pigment-producing organisms in the oral cavity as well as to exposure of the tongue to oxidizing

agents, excessive use of tobacco, antibiotic therapy, exposure to food colorings, vitamin deficiency, gastrointestinal disorders, and poor oral hygiene. In contrast to PFPT, black hairy tongue involves the filiform papillae<sup>10</sup>.

The pathogenesis of PFPT is unclear, but hyperfunction of melanocytes can be thought as a possible one because the melanin granules in melanophages are believed to be derived from melanocytes in the mucous membrane<sup>3</sup>. Hyperfunction of melanocytes is also present in other oral pigmentary disorders such as oral melanoma, melanocytic nevi, and oral melanotic macule<sup>11</sup>. In the case of oral melanoma, there are two different types of presentation, one in which a nodule is noted to be rapidly growing and the other in which pigmented macules are noted to enlarge and spread gradually before invasive growth occurs<sup>12</sup>. Although the correlation between hyperfunction of melanocytes and malignant transformation of them has not been clearly defined, pigmented macules with an uninformative history or any atypical appearance should be thoroughly evaluated with incisional or excisional biopsy. Those with a reliable history of a long-standing lesion without clinical alterations could be safely observed without invasive histopathologic examination.

To the best of our knowledge, there has been no report of malignant change in PFPT. Clinical and histopathologic examination makes it possible to discriminate PFPT from malignant oral pigmentary disorders. Once the entity of PFPT is recognized, the clinician need not concern himself or his patient with a long differential of ominous diagnoses<sup>2</sup>. Additional observations and investigations of PFPT might help to further elucidate the pathogenesis of lingual hyperpigmentation.

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