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CASE REPORT

Solitary pigmentation of the tongue: lentigo simplex or pigmented fungiform papilla?

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Abstract

The report discusses a solitary pigmentation on the dorsal tongue of an adult female, featuring variably concentrated melanocytes, hyperpigmened keratinocytes and pigmentary incontinence spatially related to fungiform and adjacent filiform papillae. Interpretations included lentigo simplex and pigmented fungiform papilla, and are critically discussed in conjunction with clinical considerations.

Introduction

While melanocytic naevi and melanotic macule of the oral mucosa are established[1–4], little or no attention has been paid to lesions of intermediate pathology therein. In addition, pigmented fungiform papillae of the tongue have been repeatedly explored by dermatologists[5,6], but are not widely known among specialists in oral disease[7]. In this context, the following case of a solitary pigmentation of the tongue, which showed features reconcilable with pigmented fungiform papilla or lentigo simplex, a lesion possibly spanning the range between naevi and melanotic macule, is of interest.

Case report

A 32-year-old, Caucasian female presented to the Oral Medicine Clinic, complaining of a dark patch on the top of her tongue. The patient was 9 months post-partum at presentation, asymptomatic and generally well. She had been aware of the patch for around 2–3 years and reported it had become more noticeable during her pregnancy. The medical history was unremarkable and the patient took no medications.

On clinical examination, an isolated 6.0 mm oval area of brown pigmentation was noted on the left dorsal surface of the tongue (Fig. 1). The area did not appear raised, had reasonably well-defined borders, did not blanch with pressure and was soft on palpation. The mucosal surface was intact, with no ulceration or contact bleeding. The patient reported no other areas of pigmentation on her body.

The clinical impression was of a melanotic macule. An incisional biopsy under local anaesthesia was performed to establish a definite diagnosis. Healing was uneventful.

Histological examination of the routinely preserved and processed material obtained during biopsy, showed lingual mucosa and underlying musculature. The former included a fungiform and a few filiform papillae. The stroma supporting the fungiform and adjacent filiform papillae showed prominent melanophages (Fig. 2A) decorated on special

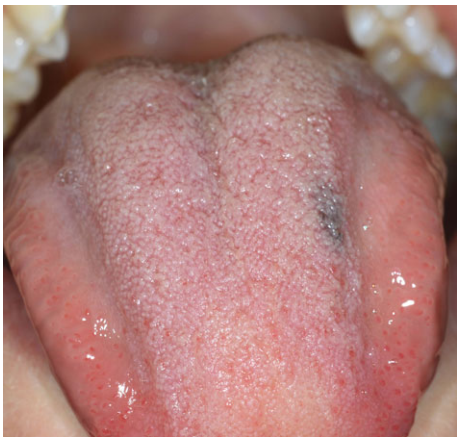

Figure 1 Lesion at presentation.

staining (Fig. 2B), which also accentuated variable hyperpigmentation of overlying deep keratinocytes (Fig. 2C). Immunohistochemistry for Melan-A and S-100 protein with the use of alkaline phosphatase instead of peroxidase revealed variable concentration of intraepithelial melanocytes in those areas (Fig. 2D). Melanocytes were not seen in adjacent surface epithelium/filiform papillae (Fig. 2E).

Discussion

The histological findings of hyperpigmentation of basal keratinocytes and melanophages in the lamina propria (pigmentary incontinence) are within the range of oral melanotic macule[4]. This would correspond with the clinical suspicion. However, melanotic macules are rare (3.3%) on the tongue and lack increased melanocytic numbers[4,8,9]. Such an increase together with keratinocytic hyperpigmentation and pigmentary incontinence would tip the scales towards a diagnosis of lentigo simplex[8–10]. Increased melanocytes featured in the present case, particularly in terms of their localised concentration and absence from adjacent epithelium (Fig. 2D, E). Paucity or even absence of melanocytes from the human tongue has been reported, but is controversial (see Barrett and Scully for a review and references[11]). Allowing for degrees of plausibility, the present findings may reflect lentigo simplex, their papillary spatial relationships being incidental to the concentration of fungiform papillae in the anterior tongue[12] and widespread abundance of dorsal filiform papillae.

The alternative interpretation would be pigmented fungiform papilla of the tongue (PFPT). It has been noted that PFTP is not alien to the medical literature. Holzwanger *et al.* discussed interesting historical aspects and reported that, of 200 African-Americans examined, 30% of females and 25% of males showed PFPT; the corresponding percentages for 100 Caucasians were 2% and 0% respectively[5]. A later investigation of 14 346 Chinese dermatological outpatients recorded 58 (0.4%) PFPT cases (56 females and 2 males)[6]. Reports of PFPT in other ethnic groups, for instance African[13], South Asian[14,15], Brazilian[16], Hispanic[5,7], Indian[17], Korean[18] and Middle Eastern[19], are sporadic. Adibi *et al.* felt that the dearth of pertinent dental publications is remarkable, and proposed 'papillary tip melanosis' as a more appropriate term[7]. We share the feelings of those authors, but do not support changing terminology because of inevitable confusion. Holzwanger *et al.* also suggested three clinical patterns of PFPT: type I refers to a single area of pigmentation, with clustering of involved papillae; type II refers to the involvement of between three and seven papillae scattered around the tongue; and type III refers to the rare case in which all fungiform papillae are involved[5]. Type II was more common in the sample examined by those authors[5], but the Chinese investigation indicated type I[6]. Our case could qualify as type I PFPT.

Before hailing the present clinicopathological presentation as PFPT in a Caucasian, the following comments are deemed necessary. The rather diffuse background pigmentation in our case (Fig. 1) contrasts with the 'punctuate' pattern in previously published clinical pictures of PFPT[5–7, 13–19]. This may be attributable to spreading of changes in an adjacent filiform papilla (Fig. 2B), a feature also noted by Adibi *et al.*[7]. Light-photomicrographs of PFPT have been published. They are usually focussing on melanophages in the lamina propria[5–7, 18] and, occasionally, keratinocytic hyperpigmentation[7]. In contrast with our observations, melanocytes are ignored, though a 'clear' intraepithelial cell, possibly a melanocyte, is discernible in a photomicrograph given by Holzwanger *et al.*[5].

The above interpretations are not mutually exclusive. Unless of 'clear' appearance, identifying and counting melanocytes is difficult in areas of keratinocytic hyperpigmentation. Likewise, conventional immunohistochemistry for melanocytic markers would be unhelpful as immunoperoxidase labelling yields brown reaction product indistinguishable from melanin. On the other hand, immune-alkaline phosphatase procedures, as applied here, yield a red reaction product and reveal melanocytes in colour contrasting with melanin. This would allow

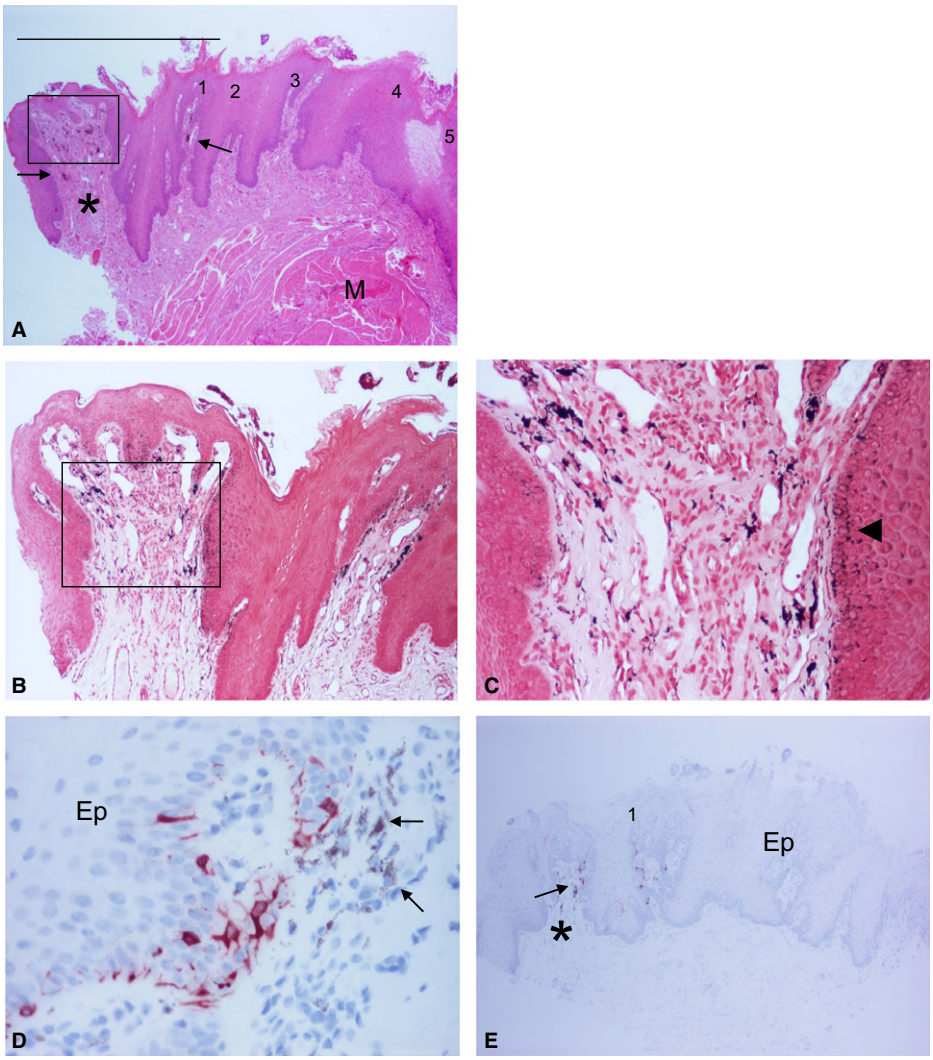

Figure 2 (A) Scanning view of section stained with haematoxylin and eosin (objective magnification × 2) shows fungiform (asterisk) and at least five filiform papillae (1–5); pigmented elements in papillary stroma are arrowed and confined to an area that measured 0.6 mm in length and corresponds to the linear segment; M, skeletal muscle. (B) Adjacent section stained with Masson–Fontana; pigmented elements staining black are more prominent subepithelially (objective magnification ×4). The rectangled area is magnified in (C); staining is associated with stromal melanophages and melanin-laden keratinocytes (arrowhead) (objective magnification ×10). (D) Dendritic basal melanocytes staining red, concentrate at tips of epithelial (Ep) rete; melanophages (arrows) are unstained (Melan-A, objective magnification ×20). (E). Scanning view of immunohistochemically treated section shows localisation of stained melanocytes in the fungiform (asterisk) and contiguous filiform (1) papillae, whereas adjacent epithelium (Ep) and melanophages (arrow) are unstained (compare with Fig. 2A) (Melan-A, objective magnification ×2).

confident identification and together with resolving uncertainties about lingual melanocytes[11], achieve a desirable terminological and conceptual precision.

Turning to aetiological considerations, sex hormones may have a role in the development of PFPT[6]. This is supported by the increased occurrence of PFPT in females[5,6], and its association with Hori's nevus and melasma, which express higher levels of oestrogen receptors[6]. Isolated observations also suggest a drug-induced mechanism[7]. Interestingly, our patient noticed the lesion became more prominent during pregnancy, and was not on medication. The suggestions do not, however, explain the spatial relationship between incontinent melanocytes[20] and fungiform papilla, which is illustrated in Fig. 2E. That this may be incidental has been discussed, but speculation over biological events is tempting. Fungiform papillae in mammals are richly innervated, and possibly depend on species-related, trophic influences of the chorda tympani, lingual nerve and

cranial sympathetics for their appearance and maintenance[21–28]. Traditional views do not favour a relation between melanocytes and innervation, but nerve branches are present in dermal papillae and the overlying epidermis accommodates melanocytes[20].

The aforementioned Chinese investigation found that 76% of individuals with PFPT were in the 2nd or 3rd decades, age ranging from 4 to 83 years[6]. The wide age range together with a possible hormonal dependence and benign nature of PFPT are similar to patterns of physiological oral pigmentation[7]. Accordingly, no treatment is necessary and confident clinical recognition should preclude biopsy[7]. In this vein, dermoscopy has recently drawn attention[15,29,30], though should be probably reserved for type II and III lesions. We would advise biopsy for type I lesions of diffuse rather than punctuate pigmentation.

It is hoped that the present report highlights clinicopathological presentations frustratingly ignored in the dental literature, and the approach to various interpretations, although largely academic, would increase understanding and diagnostic skills.

Conflicts of interest

The authors have stated explicitly that there are no conflicts of interest in connection with this article.

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