Transient lingual papillitis

Ida M. Kornerup, DDS, Dip Paedo1/Mireya Senye, DDS, MSc TMD/Orofacial Pain, MSc Orthodontics, FRCS (C)2/Edmund Peters, DDS, MSc, FRCD (C)3

A case of recurrent, clinically innocuous, but painful papules involving the tongue dorsum of a 25-year-old man is presented. The lesions were interpreted to represent a transient lingual papillitis. This a poorly understood, but benign and self-limited condition involving the tongue fungiform papillae, which does not appear to be widely recognized. (Quintessence Int 2016;47:871–875; doi: 10.3290/j.qi.a36888)

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Transient lingual papillitis (TLP) is characterized by recurrent, painful, focal or multifocal, red or yellow/white papules involving the tongue dorsum, which can occur over a wide age range from children to adults.1,3 The papules represent inflamed, variably ulcerated, fungiform papilla. TLP was first formally described by Whittaker et al1 in 1996, who suggested, despite lack of earlier documentation, that this was a relatively common condition. Since this report, there have been a small number of publications that have further documented the condition.4-13 However, the pathogenesis and management considerations remain poorly defined.

Different patterns of clinical expression have been described. These include localized involvement of one or more inflamed fungiform papilla, most frequently on the anterior tongue dorsum or a more generalized involvement, primarily affecting the anterior and lateral tongue dorsum. A third, non-painful, papulokeratotic variant, characterized by multiple, recurrent, more diffusely distributed, hyperkeratotic white/yellow papules has also been described.2,6 A further non-painful condition, termed “chronic lingual papulosis,” characterized by focal or diffuse enlargement of lingual papilla has been described. In contrast to TLP, this was thought more likely to represent fibrotic enlargement of filiform papillae (rather than fungiform papillae), occurring in response to low-grade chronic irritation. The relationship of chronic lingual papulosis to TLP is uncertain. It could represent an end-stage presentation of a long standing TLP or, alternatively, a different, etiologically distinctive, condition.12

In spite of the putative common occurrence of TLP, it does not appear to be well recognized. This paper presents a case that fit the clinical profile of TLP, and discusses the relevant literature.

CASE REPORT

A 25-year-old Caucasian man presented with a complaint of painful “tongue bumps”, which had been...
intermittently developing for over 2.5 years. He indicated that between 5 to 10 painful papules would develop on the lateral tongue dorsum about once a month. He would obtain immediate profound relief by removing the papules with nail clippers. However, the lesions would spontaneously recur in about 2 to 3 weeks following this self-management. There was no previous diagnosis and because of the persistent recurrent nature of the condition, he indicated increasing concerns this might represent a cancer.

Review of local associated or eliciting factors indicated the lesions were exacerbated or possibly induced with tomato sauce. There was a history of clenching. The medical history was unremarkable. There was no history of allergy.

At the initial examination, the glossal lesions were in a quiescent phase. However, localized multifocal white slightly raised lesions were evident on the left lateral aspect of the tongue and, to a lesser extent, on the right lateral aspect of the tongue. Focal resolving ulcerations were also noted at the sites of earlier surgical self-management (Fig 1).

In spite of the clenching history, there was no evidence of lateral tongue crenations or other diffuse inflammatory change involving the anterior or lateral aspects of tongue, which could occur in response to chronic parafunctional irritation involving the tongue. There was no evidence of malocclusion, including open bite, which might suggest tongue thrusting. There was no evidence of anterior gingival inflammatory changes, which might suggest a mouth breathing habit. In this regard, the patient did not indicate any concerns regarding breathing difficulties and no breathing difficulties were noted during the course of the interview and clinical exam.

Findings from the remaining exam were unremarkable although a “lip licking” habit was noted. The lip licking was in response to a perception of dryness of the vermilion.

A smear obtained from the tongue dorsum to assess the unlikely possibility of a secondary candidal infection was negative. No further testing, including culture or biopsy, was obtained.

Empirical management included patient education to improve awareness of the possible role of factitial injury associated with oral parafunctional activity and the potential role of topical allergenic agents. The patient was encouraged to refrain from surgical self-management. Fabrication of an occlusal stabilization appliance for sleep bruxism was recommended and a 0.12% chlorhexidine solution was prescribed to sponge on affected areas to help manage possible secondary infection.

The patient presented for follow-up after 13 months about a week after an exacerbation. He had not obtained the splint but indicated that application of the chlorhexidine solution afforded mild relief within 1 or 2 days after an exacerbation and he also felt this improved resolution when he performed surgical self-management. He noted that development of the lesions occurred approximately once every 1 or 2 months and seemed to correlate with stress. He had become aware of his habit of forcefully pressing his tongue against his palate and maxillary teeth. Again, lateral tongue crenations were not noted, possibly because the parafunctional glossal stresses were not directed against mandibular teeth. Figure 2 shows an erythematous sensitive papule and recent focal lesions, which had been clipped and appeared to be developing again. Further empirical management with topical corticosteroid application to be used at inception of the lesions has proven useful to help control the development of the lesions, limit duration to about 1 day, and to moderately reduce sensitivity. However, lesions occasionally still develop after a 2-year follow-up and thus, there is not definitive resolution.

DISCUSSION

The tongue lesions described in this report were interpreted as a reactive condition, preferentially affecting fungiform papilla for unknown reasons. The condition fit the profile of a TLP and the patient was reassured that there were no features suggesting malignancy. Categorizing the case as TLP was useful to remove the stress associated with a potential cancer diagnosis and
to exclude other inappropriate diagnoses. However, TLP is still poorly defined and the diagnosis was of limited value with respect to identifying definitively an etiologic basis or in terms of directing further treatment. The evidence implicating fungiform papilla is primarily based on the clinical inference that the sensitive papules, which characterize this condition, mimic the distribution and anatomic structure of fungiform papilla. Even though this interpretation has been widely accepted and appears reasonable, we were not able to find any photomicrographs in reported cases, which showed convincing histologic features of a fungiform papilla. These features should include a thin, non- or lightly keratinized, layer of stratified squamous epithelium and the presence of taste buds.14,15

A striking feature of the present case was the marked sensitivity, which appeared completely disproportionate to the presenting clinical features. This discrepancy is further illustrated in Fig 3, which shows painful recurrent tongue lesions in a 21-year-old healthy Oriental woman. Examination, with the patient’s active assistance, confirmed that the inconspicuous white dorsal tongue papules corresponded to the painful lesions. There had been five previous acute episodes over a 2-year period, each lasting about 8 to 9 days. Previous diagnoses had included “thrush” and an unspecified viral infection. The earlier management had included nystatin and fluconazole, which were of predictably limited benefit. Subsequent empirical management with topically applied 0.2% chlorhexidine with topical corticosteroids was useful to control pain and minimize duration of the occasional recurrence to about a day in a subsequent 4-year follow-up period.

The unusual sensitivity associated with these lesions could be because fungiform papillae are architecturally more fragile than the surrounding robust, heavily keratinized filiform papillae. As previously noted, mature fungiform papillae have a thin non-keratinized stratified squamous epithelial lining, which supports taste buds. There is evidence, in a mouse model, that both epithelial and connective tissue components of taste buds are embryonically derived from neural crest.16,17 Structurally, the taste buds have external pores to permit ingress of chemical substances.18 Thus, the lining would be relatively susceptible to injurious stimuli, and the resultant inflammation with attendant edema would involve a relatively well-innervated fibrous core, which uniquely supports taste bud function. Possibly, these considerations explain the selective inflammation involving the fungiform papilla and the consequent localized sensitivity. In this regard, in those cases where the reactive change involves thickening of the epithelial lining (the papulokeratotic variant), sensitivity does not occur.

A variety of injurious pro-inflammatory stimuli or predisposing conditions have been described. Different
reports have noted that since the common end point in TLP is an inflamed fungiform papilla, there is no reason that this could not be mediated through a range of pro-inflammatory factors.\textsuperscript{1,3,7}

The suggested eliciting factors have included irritation associated with chronic trauma or parafunctional glossal activity,\textsuperscript{1,19} chemical damage described in an individual licking envelopes,\textsuperscript{16} hypersensitivity to foods or oral hygiene products,\textsuperscript{2,3,8,9} guttate psoriasis,\textsuperscript{21} and hormonal predisposition.\textsuperscript{2} The possibility of viral infection has also been suggested in a similar condition termed “eruptive lingual papillitis”.\textsuperscript{4,7} These reports described a condition characterized by recurrent inflammation of the fungiform papillae of the tip and lateral tongue dorsum, primarily in children. There was variable association with lymphadenopathy, fever, and angular cheilitis. Possible transmission to family members was noted.

Diagnosis is made on the basis of the clinical presentation, and biopsy is generally not necessary. In the occasional cases where a biopsy is obtained, the histopathologic presentation is nonspecific. The papillae show chronic inflammation with edema, possibly with ulceration or mild hyperkeratosis. The non-painful papulokeratotic variant shows epithelial hyperplasia, hyperkeratosis, and chronic inflammation.\textsuperscript{1,2,4-6}

Clinical exclusion of other possible diagnoses is necessary and the differential diagnosis could include herpetiform aphthous stomatitis and recurrent herpetic infection.\textsuperscript{3} However, recurrent herpetic ulcers initially present as vesicles, typically on masticatory mucosa. Herpetiform aphthous ulcers are very unusual and present as multiple punctate ulcers involving erythematous mucosa, usually on non-keratinized mucosa. In both cases, these evolve into obvious ulcerations, which are distinct from TLP. Fungiform papilla enlargement has also been noted in other clinical contexts, which appear distinctive from TLP. These include asymptomatic fungiform papilla enlargement, which can occur with ciclosporin A use\textsuperscript{22} and in neurofibromatosis.\textsuperscript{23} Other systemic conditions that can show fungiform papilla pathosis include scarlet fever, familial dysautonomia, some anemias, and with vitamin deficiencies, which result in dorsal tongue atrophy.\textsuperscript{6}

Well-defined clinical management protocols have not been established and treatment remains empirical and palliative.\textsuperscript{1,3} Among the proposed treatments are use of topical anesthetics, corticosteroids, coating agents, chlorhexidine rinse, change of oral hygiene products, identification and elimination of potential allergenic agents, and control of parafunctional habits. The use of nail clippers to remove the lesions, which was practiced by our patient, has been previously documented and is not recommended.\textsuperscript{1,2} In the present cases, management with topical corticosteroids in conjunction with topical application of 0.2% nonalcoholic chlorhexidine solution has proven useful. Patient reassurance regarding the benign nature of the condition was important.

**CONCLUSION**

In summary, although there has been a suggestion that TLP is a common condition, the incidence is not known. Retrospective surveys, attempting to assess the frequency of presentation based on patient recollection, should be treated with caution. Diagnosis should be restricted to those cases in which it is obvious that there is selective involvement of fungiform papillae after examination by qualified clinicians. The condition is benign and self-limiting although often painful and recurrent. Management involves efforts to identify and eliminate any potential local or systemic factors and empirical conservative topical palliative treatment. In the present cases, patient education with reassurances regarding the benign nature of the condition was important.

**REFERENCES**