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## Correspondence

# Verruciform xanthoma of the tongue



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Oral verruciform xanthoma is an uncommon, reactive lesion of unknown etiology and is most commonly found on the gingiva.<sup>1</sup> Here, we reported a case of verruciform xanthoma presenting as a white slightly papillary lesion on the right posterior ventral surface of the tongue in a 69-year-old female patient.

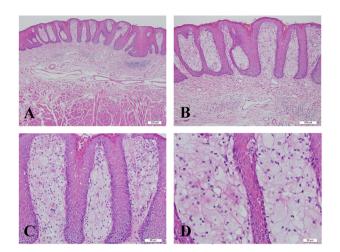
This 69-year-old female patient came to our dental clinic for evaluation and treatment of a white slightly papillary lesion on the right posterior ventral surface of the tongue for more than one month. The lesion was a slightly elevated plague measuring approximately 1.0 cm in greatest dimension. It is asymptomatic and not tender on palpation. The clinical diagnosis was a papillary lesion or an oral leukoplakia. After discussing with the patient and obtaining the signed informed consent, the lesion was totally excised under local anesthesia. The removed soft tissue specimen was sent for histopathological examination. Microscopically, it showed parakeratosis of the surface stratified squamous epithelium with a slightly papillary surface. The epithelial ridges were elongated and of uniform depth. There was a mild to moderate lymphoplasma cell infiltrate in the lamina propria and submucosa (Fig. 1A and B). The most characteristic finding was the accumulation of lipid-laden foamy macrophages in the connective tissue papillae between epithelial ridges (Fig. 1C and D). The above-mentioned characteristic findings finally confirmed the histopathological diagnosis of an oral verruciform xanthoma.

The vertuciform xanthoma is so-called because it often has a papillary or vertuciform surface and the lipid-laden foamy macrophages are usually designated as xanthoma cells.<sup>1-3</sup> A largest series of 212 cases of oral vertuciform xantoma has been reported by Belknap et al.<sup>1</sup> The mean age of the 212 patients was 61 (range, 9 to 94) years. These lesions occurred nearly evenly in male and female patients with a female to male ratio of 1.06: 1. The most common location of oral verruciform xanthoma was the gingiva (n = 110), followed by the palate (n = 41), tongue (n = 20), buccal mucosa (n = 18), vestibule (n = 13), lip (n = 4), floor of mouth (n = 3), and unspecified site (n = 1) in a descending order. Three of the 212 reported cases were recurrent lesions.

The etiology and pathogenesis of oral verruciform xanthoma remains unclear.<sup>1</sup> However, two main hypotheses have been proposed in the literature. Zegarelli et al.<sup>2</sup> suggested that the accumulation of lipid-laden macrophages may result from epithelial cell degeneration with release of the lipid material, which is scavenged by the macrophages. Nowparast et al.<sup>3</sup> suggested that the verrucous and papillary epithelial architecture seen in oral verruciform xanthoma is probably secondary to the presence of foamy histiocytes, which affect the metabolism of the epithelial cells, leading to a hyperkeratotic epithelial surface. Both hypotheses support a mutual interaction among macrophages, keratinocytes, and chronic inflammation. We also found a significant chronic inflammatory cell infiltrate in the subepithelial connective tissue of our oral verruciform xanthoma lesion, which supports the inflammatory hypothesis as the main etiology in the development of oral verruciform xanthoma. Immunostain is useful for identification of tumor cell origin.<sup>4,5</sup> By immunostain, the lipid-laden macrophages are positive for CD68 (a macrophage marker) as well as positive for CD63 and CD163 (both are monocyte/macrophage markers), but are negative for \$100, CK8, CK19, CK14, and pancytokeratin.<sup>1</sup>

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**Figure 1** Histopathological microphotographs of our case of oral verruciform xanthoma. (A and B) Low- and medium-power microphotographs showing parakeratosis of the surface stratified squamous epithelium with a slightly papillary surface and elongated epithelial ridges of uniform depth. There was a mild to moderate lymphoplasma cell infiltrate in the lamina propria and submucosa. (C and D) High-power microphotographs demonstrating a characteristic finding of accumulation of a sheet of lipid-laden foamy macrophages in the connective tissue papillae between epithelial ridges. (Hematoxylin and eosin stain; original magnification; A,  $4\times$ ; B,  $10\times$ ; C,  $20\times$ ; and D,  $40\times$ ).

## Declaration of competing interest

The authors have no conflicts of interest relevant to this article.

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