CASE REPORT

Oral verruciform xanthoma

Lydia Harris,¹ Konrad Staines,² Miranda Pring³¹Oral & Maxillofacial Department, Royal United Hospital Bath NHS Trust, Bristol, UK²Department of Oral Medicine, Bristol Dental Hospital, Bristol, UK³Department of Pathology, Bristol Dental Hospital, Bristol, UK

Correspondence to

Dr Lydia Harris,
lmjharris@hotmail.co.uk

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SUMMARY

Verruciform xanthoma (VX) of the oral cavity is a benign mucosal growth that often presents as a pink, yellow or grey raised plaque or papule with granular, papillary or verrucous surface morphology. Intraorally this often presents on the masticatory mucosa and extraorally often involves the skin and anogenital mucosa. There are several proposed aetiological factors and the clinical features of VX can be misleading; clinically it can resemble malignancy. Histopathological diagnosis is a key for the correct management of this lesion. Excision of this lesion is curative.

BACKGROUND

This clinicopathological case details an unusual presentation of verruciform xanthoma (VX) arising in the floor of the mouth. Its aetiology, common histopathological features and management are also discussed.

This case is important due to its misleading clinical appearance and interesting histopathology, and highlights the need for further investigation.

CASE PRESENTATION

A 45-year-old patient presented to the oral medicine department with a 2-month history of a painless lump in the floor of the mouth. The patient had no relevant medical history and was a non-smoker. Extraoral examination showed no obvious abnormality. Intraoral examination revealed a well-demarcated white plaque measuring 7 mm in diameter. This was centred on the left ventral surface of the tongue/floor of the mouth and demonstrated a verrucous appearance with no evidence of ulceration or significant erythema. It was firm on palpation, slightly raised and thickened.

INVESTIGATIONS

Clinical photographs were taken and an incisional biopsy was performed (figure 1A, B).

Histopathologically, the biopsy demonstrated mucosa with evidence of surface papillomatous architecture. The epithelium was hyperplastic with prominent surface parakeratosis and broad elongate rete ridges. The presence of foamy macrophages was noted immediately beneath the squamous epithelium as aggregates (figure 2A, B). This was confirmed as CD68 positive macrophages were present at the epithelial/connective tissue interface but these did not extend below the level of the adjacent rete pegs (figure 2B, C). There was no epithelial dysplasia and a D/PAS stain for fungi was negative.

Figure 1 (A) Sublingual verrucous swelling with scale. (B) Sublingual verrucous swelling.

DIFFERENTIAL DIAGNOSIS

Prior to incisional biopsy being undertaken, the clinical differential diagnosis included that of malignancy (squamous cell carcinoma, verrucous carcinoma) or squamous papilloma. The microscopy (above) revealed the presence of foam cells and characteristic histological features of verruciform xanthoma.

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Figure 2 (A) H&E stained section, original magnification $\times 80$. (B) H&E stained section, original magnification $\times 200$. (C) Immunohistochemistry CD68, original magnification $\times 100$.

Histologically, the differential diagnoses included those related to the squamous proliferation (eg, squamous dysplasia, human papillomavirus, squamous cell carcinoma, fungal infections) and diagnoses related to the foamy macrophages (xanthoma, VX,

granulomatous conditions, eg, sarcoidosis). This case was thought to be typical for VX given the combination of appearances.

TREATMENT

VX is a benign mucosal swelling and excision is usually curative, although recurrence has, rarely, been reported.¹

OUTCOME AND FOLLOW-UP

Following incisional biopsy confirming the diagnosis, the lesion was excised and following a 6-month review period there has been no recurrence.

DISCUSSION

VX of the oral cavity is a benign mucosal growth presenting most commonly in the masticatory mucosa, in particular at the gingival margin. It is a rare finding in the floor of the mouth.^{1 2} It often presents as a pink, yellow or grey raised plaque or papule with granular, papillary or verrucous surface morphology.^{3 4} It is usually asymptomatic and slow growing, and more often occurs as a solitary lesion, although multiple lesions have occasionally been reported.^{1 5} VX can also present extraorally, in particular on the skin and anogenital mucosa. VX is slightly more frequently seen in men and Caucasians, and has been associated with other oral conditions including oral pemphigus vulgaris, dysplasia and lichen planus.⁴

The differential diagnosis is important as it can clinically resemble malignancy. Differentials include verrucous carcinoma, squamous cell carcinoma, squamous papilloma, verruca vulgaris, condyloma and leukoplakia.^{4 5} The verrucous architecture and exophytic nature of this lesion could easily mimic malignancy clinically and therefore biopsy is mandatory. A reassuring clinical feature is that these lesions tend to be slightly softer on palpation, unlike a malignant process such as verrucous carcinoma.

The aetiology of VX is largely unknown, although a myriad of aetiological factors have been implicated. It has been attributed to trauma and inflammation, as it is more commonly seen in masticatory mucosa. An immunological cause has been suggested due to the presence of Langerhan cells within the lesions.² It has also been proposed that epithelial entrapment with degeneration and accumulation of lipids leads to the formation of xanthoma cells or foam cells,⁶ or that the pathogenesis involves excessive accumulation of lipids, which are then gathered by macrophages.²

The presence of foam cells is characteristic of VX; these tend to accumulate in between the rete ridges in the connective tissue. The squamous epithelium is usually hyperparakeratinised and often presents with a papillary architecture. One pitfall is that hyperplastic papillary projections may present with cross cut artefacts resulting in a pseudoinvasive appearance and risk of a histological misdiagnosis of a malignancy.

Learning points

- ▶ Oral verruciform xanthoma (VX) is a largely asymptomatic lesion more commonly seen on the masticatory mucosa.
- ▶ The differential diagnosis includes verrucous carcinoma, squamous cell carcinoma, squamous papilloma, verruca vulgaris, condyloma and leukoplakia.
- ▶ Clinically, VX can resemble malignancy therefore biopsy is mandatory.
- ▶ The diagnosis is based on histopathological findings.
- ▶ Curative treatment is localised surgical excision.

Contributors LH, KS and MP were involved in the management and diagnosis of the patient. LH authored the article with input from KS and MP.

Competing interests None.

Patient consent Obtained.

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