

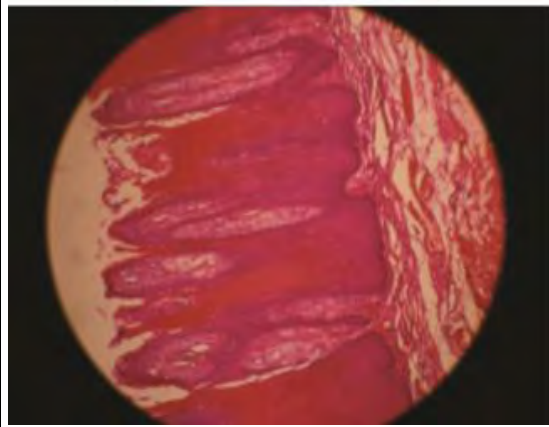
原文題目(出處)：	Oral verruciform xanthoma: A case report and literature review. Case Rep Dent, Article ID 528967
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報告日期：	103/1/14

### 一、Introduction

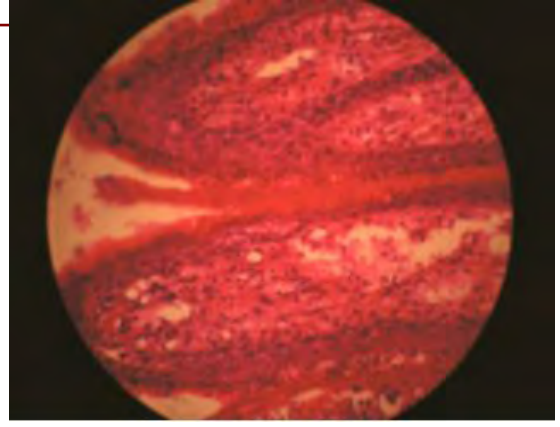
1. Verruciform xanthoma is benign muco-cutaneous lesion that resembles virus-induced papilloma but has an unknown etiology and uncertain nature.
2. It most commonly presents with a verrucous appearance. It also may appear polypoid, papillomatous, or sessile.
3. It occurs as a small (0.2–2cm), solitary, asymptomatic, slow growing, white or yellowish red lesion with no sex predilection.
4. Histologically VX is distinguished from other lesions by presence of large numbers of foam cells, and limited to the connective tissue papillae. The foam cells on ultrastructural studies have been concluded to be fat-laden macrophages. Other cell types, including Langerhans cells, intraepithelial neutrophilic infiltrate, and even fibroblasts.
5. Variable degree of parakeratosis in the crypts between papillae which are of variable length and thickness, often extending close to the surface. The rete pegs are extremely elongated and uniform.
6. Almost every VX case is diagnosed on histological examination as the clinical appearance is not diagnostic. Differentiation from other lesions with foamy or granular cells is not difficult because VX is only lesion has these cells confined to the papillae.
7. Differential diagnosis: erythroplasia of Queyrat (Bowen disease of the glans penis), seborrheic keratosis, verrucous carcinoma, verruca simplex, and condyloma acuminatum.
8. Treatment: local surgical excision which is almost always curative.
9. Recurrence is rare.

### 二、Case report

1. Age/sex: 60-year-old male
2. Chief complaint: a painless growth on the left buccal mucosa.
3. History: He had a habit of smoking for the past 10 years. No systemic diseases
4. On clinical examination,
  - (1) A yellowish white exophytic lesion, measuring 11×7 mm in size, which had a cauliflower shape on inspection.
  - (2) The lesion was asymptomatic and soft in consistency.



Showing epithelial hyperplasia with parakeratosis and elongated rete pegs. There is an abundance of foam cells in the connective tissue papilla (H&E stain 10x).



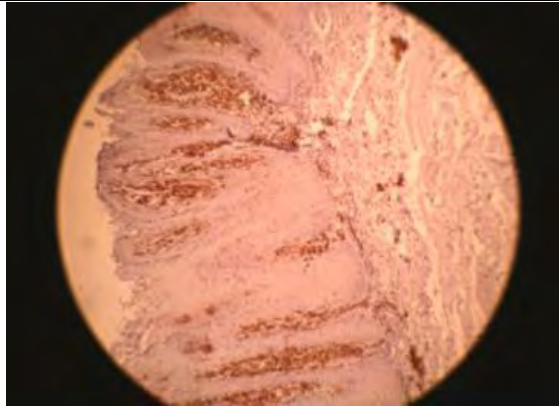
Presence of large numbers of lipid-laden foamy histiocytes confined to the connective tissue papillae (H&E stain 40x).

- (3) Lymph nodes were not palpable.
- (4) A provisional diagnosis of papilloma was made. After clinical examination an excisional biopsy was taken to rule out malignancy.

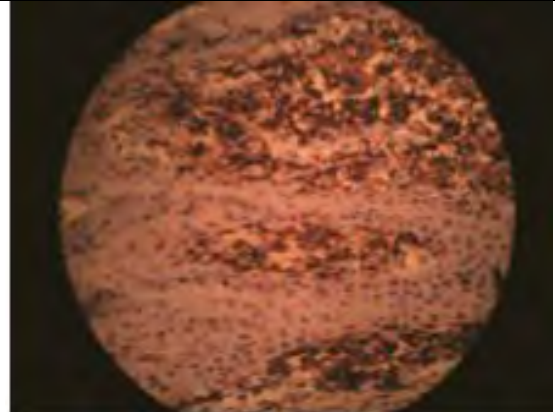


Clinical photograph showing the exophytic lesion on the left buccal mucosa.

- 5. On histopathological examination,
  - (1) Hematoxylin and eosin (H&E) stained sections showed parakeratotic epithelium with columns of parakeratin plugging into it.
  - (2) Uniformly elongated rete pegs, deep connective tissue papillae were seen, some of which extended into overlying surface epithelium
  - (3) The papillary zone of lamina propria showed numerous lipid-laden foam cells.
  - (4) There was no evidence of dysplasia or malignancy. The fact that the foam cells were confined to the papillary region of connective tissue confirmed the diagnosis as VX.
- 6. The immunohistochemical staining for CD68 was positive for the foamy macrophages. All the foam cells were strongly stained with anti-macrophage antibodies.

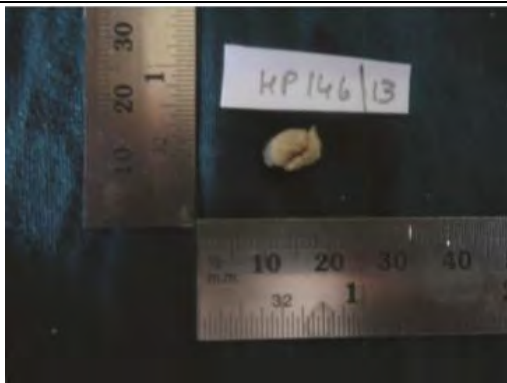


Showing foam cells with strong cytoplasmic CD68 immunostaining. The epithelial cells were negative (IHC stain 10x).



The infiltrated foam cells in the papillary dermis were CD68 positive (IHC stain 40x).

7. The treatment was performed under local anesthesia. The whole lesion was excised in one piece during excisional biopsy. Postoperative check-up showed no sign of recurrence.



Showing the lesion after excision.

### 三、Discussion

1. Verruciform xanthoma is a rare lesion, accounting for 0.025– 0.095% of all cases with an unknown etiopathology.
2. It can be because of the damage to the squamous cells due to trauma, irritation, or infection, which can cause increased epithelial turn over leading to the disease.
3. The epithelial breakdown leads to an inflammatory response and a subsequent release of lipid material from the degenerated cells.
4. Most of the cases occur in otherwise healthy individuals. Due to its clinical and histopathological resemblance to human papilloma virus(HPV)-induced lesions, verruciform xanthoma was believed to be caused by HPV. However, most investigators have not found any evidence for the presence of HPV in these lesions.
5. A few cases have been reported associated with inflammatory conditions such as pemphigus vulgaris, lichen planus, discoid lupus erythematosus, warty dyskeratoma, epidermal nevus/CHILD nevus, dystrophic epidermolysis bullosa, and seborrheic keratosis. A few cases have also been reported to be associated with disorders of lipid metabolism.

6. On serological examination(血清學檢查), our case had a normal serum lipid profile.
7. The lesion is benign, non-symptomatic, and slow growing and rarely exceeds 2cm in size. It is sessile or pedunculated and can resemble leukoplakia or squamous papilloma.
8. It occurs most commonly in 4th–6<sup>th</sup> decade of life with equal distribution between both sexes. However, it has been reported that there is a slight male predilection.
9. The lesion is usually present in intraoral regions especially on alveolar ridge, gingiva, followed by buccal mucosa, palate, floor of the mouth, and lip. Extra oral sites include vulva(外陰) , , scrotum(陰囊), penis, and skin of thigh and perineum(會陰), which are usually associated with other conditions like lymphedema(淋巴水腫), epidermal nevi, congenital hemidysplasia, and limb defect syndrome .
10. Histologically, VX shows three patterns: verrucous (most common), flat , and papillary(least common).
  - (1) There is parakeratosis of the hyperplastic epithelium which is variable in extent and is usually more marked in the verrucous and papillary patterns.
  - (2) The rete pegs are elongated, uniform, and thin, with deep central keratinized clefts and keratin plugging.
  - (3) There is no evidence of dysplasia. The characteristic histological feature is the presence of xanthoma cells in the connective tissue. There is a controversy over the exact origin of these cells. They are said to be a lineage of monocytes/macrophages .The lipid found in the xanthoma cells is said to be the same as seen in other inflammatory reactions.
11. In our case, the foam cells showed strong CD68 immunoreactivity. CD68 is a cell marker confirming the possible role of macrophages in the formation of foam cells.
12. The treatment of choice is complete surgical excision which is very effective with no recurrence. However(例外), a recurrent VX of the vulva has been reported in a 30-year-old woman, 8 years after the initial treatment . Histopathological examination of the biopsies should be performed to distinguish VX from other verrucous lesions.

題號	題目
1	下列關於Verruciform xanthoma 的敘述何者為非? (A) 屬於一種autosomal dominant disease (B) 通常沒有明顯臨床症狀 (C) 和HPV的感染無關 (D) 一般治療方式是採取complete surgical excision,復發率低
答案(A)	出處：Oral and Maxillofacial Pathology (3 <sup>rd</sup> edition) p.1020

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題號	題目
2	下列疾病皆與HPV的感染有關聯,下列那一選項最無關?
	(A) Squamous cell carcinoma (B) Squamous papilloma (C) Condyloma Acuminatum (D) Verruciform xanthoma
答案(D)	出處：Oral and Maxillofacial Pathology (3 <sup>rd</sup> edition) p.1015