

# Oral condyloma acuminatum with changes in excretory duct of minor salivary gland: An unusual case report and review of literature

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## Abstract

Condyloma acuminatum (CA) is human papilloma virus-induced disease, rarely involving oral cavity, usually sexually transmitted and frequently occurs in anogenital areas. Oral lesions are being present predominantly on the tongue, lip mucosa, buccal mucosa, palate and floor of the mouth. The disease is more common in teenagers and appears as solitary or multiple, sessile or pedunculated masses. Here, we report the unique case of CA on the lower lip in a 45-year-old man, showing changes in the excretory duct of the minor salivary gland, which is relatively unusual.

**Keywords:** Condyloma acuminatum, human papillomavirus, oral

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## INTRODUCTION

Condyloma acuminatum (CA) caused by human papillomavirus (HPV), is often considered to be a sexually transmitted disease and frequently occurs in anogenital areas.<sup>[1]</sup> Oral CA is an increasingly common but frequently undiagnosed disease. CA generally appears 1–3 months after exposure to an infected partner and presents in multiple forms in the oral cavity.<sup>[2]</sup> Oral CA was first published by Knapp and Uohara in 1967.<sup>[1,3]</sup> CA is usually diagnosed in teenagers and young adults, but all ages are susceptible. Oral lesions often involve labial mucosa, lingual frenum, tongue, buccal mucosa, floor of the mouth and soft palate.<sup>[4,5]</sup> Oral lesions can occur without accompanying anogenital lesions.<sup>[3]</sup> CA is the most commonly associated with low-risk type of HPV including HPV 6 and 11.<sup>[6]</sup> Oral

CA may involve the excretory ducts of the minor salivary gland.<sup>[7]</sup> Here, we present a case of oral CA on lower lip with changes in the excretory duct of the minor salivary gland which is relatively unusual and not much described.

## CASE REPORT

A 45-year-old man presented to our hospital with a 6-month history of a growth on his right side lower lip. On examination, three sessile nonulcerated grayish-white, soft, nontender raised circumscribed lesions were present on the right side lower lip, extending up to the commissure [Figure 1]. The family history of his wife being treated for anogenital warts was given.

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**Figure 1:** Clinical image shows multiple nodular grayish-white lesions on the right side lower lip

The largest nodule appeared to be coalescing with one of the smaller lesions, and a small satellite lesion was on the mesial side of the largest lesion. No other oral and genital lesion or cervical lymphadenopathy was noted. Serologic tests for syphilis and a test for human immunodeficiency virus were negative.

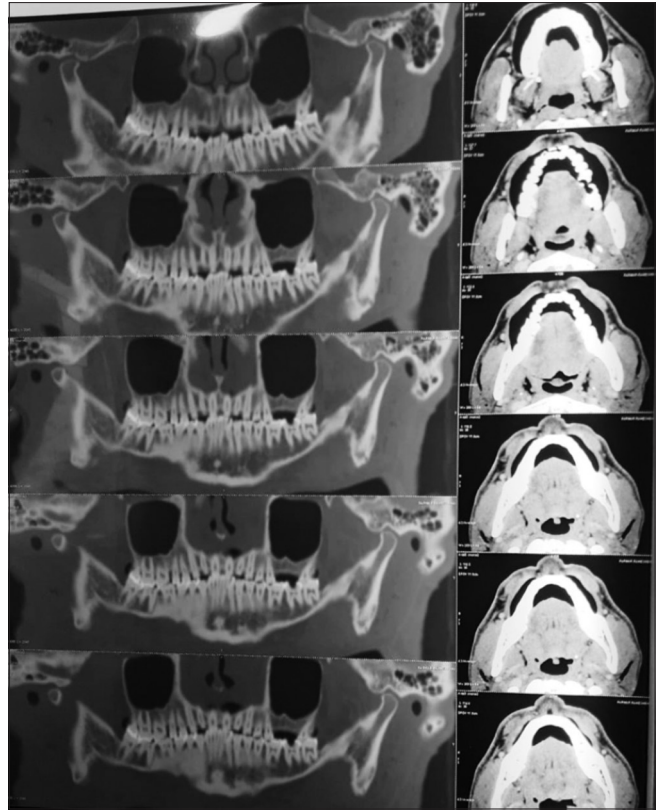
Radiographic examination revealed dentulous maxilla and mandible with normal bony trabeculation and no intrabony pathologies other than few periapical radiolucencies in the mandible [Figure 2]. Two excisional biopsy samples were obtained [Figure 3]. A wedge excision with primary closure was performed [Figure 4].

Histopathology of the excised specimen showed parakeratotic papillary proliferation of squamous epithelium with prominent acanthosis [Figure 5]. Koilocytes were seen consisting of nonuniform perinuclear halos, more prominent toward the surface [Figure 6]. The underlying connective tissue was fibrovascular consisting of dilated and congested vessels and minor salivary gland acini. Mucous and oncocytic metaplasia of the excretory duct was noted in this case [Figures 5 and 7]. All the histopathological features and clinical features were consistent with the diagnosis of CA.

Postoperatively, the patient remained free of symptoms and disease at 9-month follow-up.

## DISCUSSION

Oral CA is mostly a sexually transmitted infection, many times occurring with simultaneous genital warts in the patient or the patient's sexual partner. Nonsexual transmission has also been reported, including autoinoculation from anogenital lesions, contaminated environmental surfaces



**Figure 2:** Computed tomography scan image showing dentulous maxilla and mandible with normal bony trabeculae

and perinatal transmission from mother to newborn. Trauma may play a role in development of the lesions.<sup>[1]</sup> In the present case, the patient's wife was treated for anogenital warts.

CA presents clinical and pathological similarities to other epithelial lesions caused by HPV. Numerous HPV types have been detected in benign and premalignant and malignant lesions. Differential diagnosis of CA includes squamous papilloma, verruca vulgaris and focal epithelial hyperplasia. Verruca vulgaris is induced commonly by HPV 2, 4, 40 and 57, while CA and squamous papilloma are associated commonly with HPV 6 and 11, and HPV 13 and 32 commonly cause focal epithelial hyperplasia.<sup>[6,8,9]</sup> CA is usually diagnosed in teenagers and young adults, but all ages are susceptible.<sup>[4]</sup> The lesions are likely to be multiple and frequently occur on the tongue, lips, palate buccal mucosa and floor of the mouth or at the site of contact/due to trauma, with equal gender predilection.<sup>[4,5,10]</sup> The present case is in accordance with the literature involving lower lip and with multiple sessile lesions.

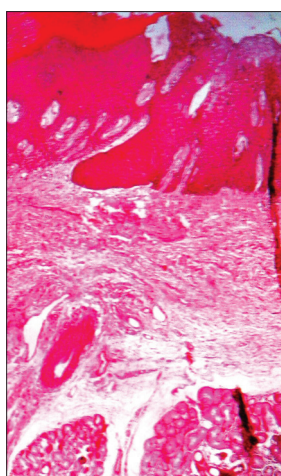
Squamous papilloma differs from CA clinically as soft pedunculated mass with numerous finger-like surface projections, whereas CA is usually sessile and seldom pedunculated.



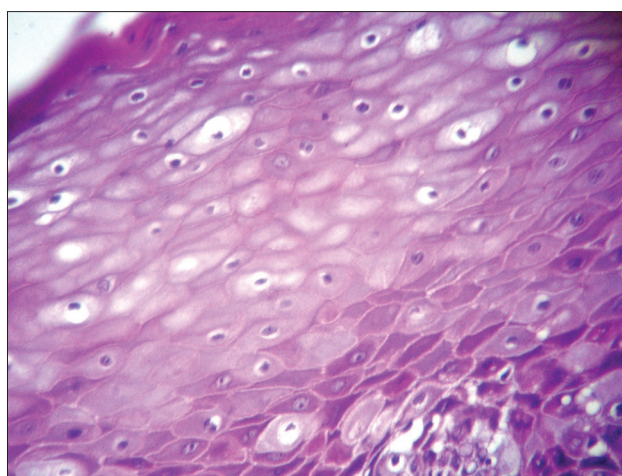
**Figure 3:** Gross specimen having pebbled surface



**Figure 4:** Clinical image postoperatively



**Figure 5:** Histopathological image showing parakeratotic stratified squamous epithelium with acanthosis. Underlying connective tissue showing excretory duct metaplasia (H&E, x40)



**Figure 6:** Histopathological image showing koilocytes with nonuniform perinuclear halos (H&E, x400)

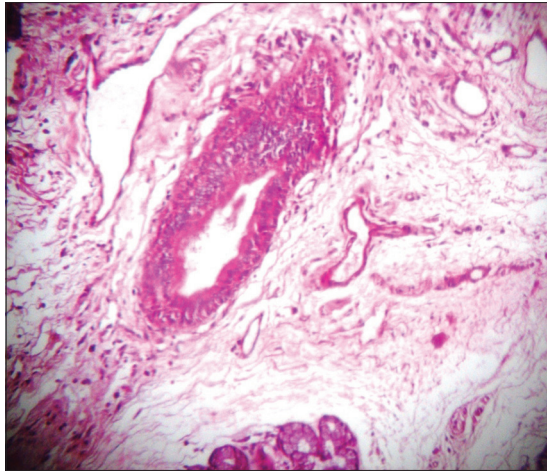
Verruca vulgaris tends to have pointed or verruciform surface projections, very narrow stalk and present as multiple or clustered individual lesions. Both verruca vulgaris and papilloma characterized by proliferation of hyperkeratotic stratified squamous epithelium arranged into finger-like projections, each with connective tissue core which is absent in CA.<sup>[4]</sup>

Focal epithelial hyperplasia (Heck's disease) lesions are broad-based or slightly elevated, smooth-surfaced, flat-topped and well-demarcated plaques. This is easily differentiated from squamous papilloma, verruca vulgaris and CA by lack of pronounced surface projections and presence of mitosoid cells.<sup>[4,6]</sup>

Histologic features, differentiating oral CA from others, are papillomatosis with prominent acanthosis and parakeratin that line deep crypts and basal cell hyperplasia.<sup>[5,11]</sup> Koilocytes, a hallmark of CA and HPV infection, are modified

keratinocytes consisting of nonuniform perinuclear halos and accompanied by nuclear enlargement and hyperchromasia.<sup>[5-7]</sup> The papillomatosis and nonuniform perinuclear halos were significantly correlated with HPV and in equivocal cases of oral condyloma.<sup>[5]</sup> In the present case, microscopic examination shows papillary parakeratotic stratified squamous epithelium with cryptic invagination in few areas along acanthosis and basal cell hyperplasia. Koilocytes with nonuniform perinuclear halos are also present in our case.

In oral CA, involvement of the excretory ducts of the minor salivary gland is previously undescribed. With the predilection of HPV infection, involvement of epithelium and extension into the ducts is exceptional but may occur.<sup>[7]</sup> Simultaneous presence of oral inverted ductal papilloma and 2 CA cases on lips is described in middle-aged HIV+ women. Both inverted ductal papilloma and 2 CA were positive for HPV 11.<sup>[12]</sup> The association



**Figure 7:** Histopathological image showing mucous and oncocytic metaplasia in the excretory duct (H&E, ×100)

between CA and extension into ducts related to HPV is unclear. Although not clear, viral infection-related immunosuppression could be associated with underlying etiopathogenesis, which require further advanced studies. In the present case, the excretory duct is also showing mucous and oncocytic metaplasia which may be a precursor of future salivary gland pathologies.

The treatment of CA includes surgical excision, electrocautery, cryosurgery and laser ablation, photodynamic therapy, Mohs microsurgery, cytotoxic agents including one or more topical application of podophyllum resin, 5-fluorouracil and trichloroacetic acid.<sup>[1,2,13,14]</sup> Because CA lesions tend to recur, patients usually require ongoing follow-up and further treatment.

## CONCLUSION

We conclude that in the oral cavity, care must be taken to differentiate between squamous papilloma, CA and verruca vulgaris. Patients with oral CA should be considered for examination and evaluation for anogenital lesions. Oral CA may involve the excretory duct of the minor salivary gland which is usually undescribed in the oral cavity. Till now, only two articles have been published which suggest the association of oral CA and salivary gland duct pathologies. Finding of mucous and oncocytic metaplasia in the excretory duct of the minor salivary gland in oral CA could probably be the precursor event of salivary ductal pathologies and hence further may possibly be used as an important diagnostic and prognostic tool in pathogenesis of salivary ductal lesions associated with oral CA and its

mimics. Further investigation and workup is required to support this observation.

## Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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## Conflicts of interest

There are no conflicts of interest.

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