

CASE REPORT



Unveiling a Novel Association: Heck's Disease caused by HPV 84

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ABSTRACT

Heck's disease is a rare benign lesion of the oral mucosa that is primarily observed in female children and is caused by HPV 13 and 32. In the oral cavity, it appears as many white and skin-colored papules. Here, we report a case of Heck's illness in an adult male with HPV 84 as the causal agent. This case highlights the importance of keeping Heck's disease as a differential in the popular white lesions seen in oral cavity and use of DNA PCR in identifying the causative agent.

KEY WORDS

Focal epithelial hyperplasia; Heck's disease; Human papillomavirus; Histopathology; Polymerase Chain Reaction; HPV 84

ARTICLE HISTORY

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Introduction

Focal epithelial hyperplasia (FEH), also referred to as multifocal papilloma or Heck's disease, is a rare benign lesion of the oral mucosa that is brought on by HPV 13 and 32 [1]. This condition primarily affects children with male to female ratio of 3:4 [2]. It is identified by several tiny, white or skin-colored papules or nodules in the oral cavity, often on the tongue, outer lip, and buccal and labial mucosa, and less frequently on the gingiva and palate [3]. Here we present a case of clinically and histologically diagnosed Heck's disease in a adult patient with positive HPV DNA and PCR sequencing for HPV 84.

Case Report

A 45-year-old male patient presented to the Dermatology Outpatient Department (OPD) with tiny whitish papules over buccal mucosa of 3 months duration with foreign body sensation (Figure 1).



Figure 1. Multiple skin coloured and yellowish papules along the angles of mouth.

The patient denied the history of any topical application. The patient had no history of high-risk sexual behavior or immuno-compromised state. On examination, patient had multiple skin-colored and yellowish papules over bilateral buccal mucosa predominantly near the angle of mouth.

Differential diagnoses of Fordyce's spots and Heck's disease were considered (Figure 2).



Figure 2. Multiple skin coloured and yellowish papules in the buccal mucosa and along the angles of mouth.

A punch biopsy was taken under local anesthesia which showed koilocytic changes in the epithelial lining along with acanthosis, throughout the epidermis and mitotic bodies were also seen. HPV DNA detection and PCR sequencing of the tissue sample was done which was positive for HPV DNA genotype 84 (Figure 3 and 4).

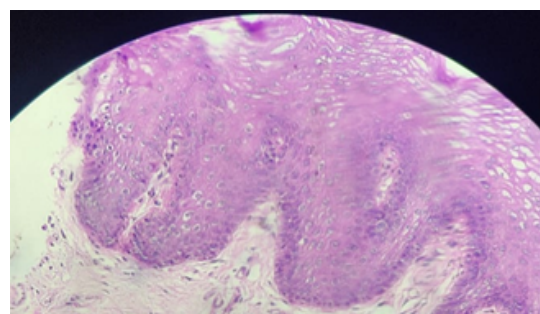


Figure 3. Photomicrograph of koilocytes and acanthosis within the epidermis (hematoxylin-eosin, original magnification x10).

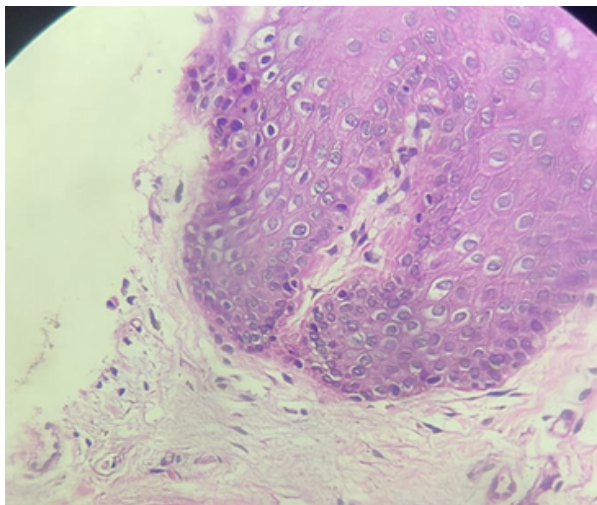


Figure 4. Photomicrograph of koilocytes within the epidermis(hematoxylin-eosin, original magnification x40).

Based on history, clinical features, histological features, and HPV DNA detection [HPV 84], a final diagnosis of FEH, Heck's disease was made.

Discussion

Human papillomavirus (HPV) causes Heck's disease, often referred to as localized epithelial hyperplasia, which is a benign lesion of the oral mucosa [4]. Archard et al. found it in Indian children in North and South America in 1965. The condition is identified by the presence of many mucosa-colored papules in the oral cavity, particularly on the tongue, outer lip, and buccal and labial mucosa [5]. Although lesions are not unusual in young and middle-aged individuals, they mostly affect youngsters [6]. The human papillomaviruses HPV-13 and HPV-32 appear to be most associated with FEH, while other HPV subtypes like HPV6, 11, 16, 18, 31, 39, 40, 51, 52, 55, 58, 66, 68, 69, 71 and 74 may be associated but are relatively rare [7]. Heck's disease may be associated with Human leukocyte antigen (HLA) DR4.

Important differential diagnoses for FEH are Fordyce spots, oral mucosal involvement of Crohn's disease, inflammatory fibrous hyperplasia, inflammatory papillary hyperplasia, Cowden's disease, condyloma accuminata and morsicatio buccarum.

Predisposing factors for FEH are hereditary predisposition, poor oral hygiene, low socioeconomic status, immunocompromised state, HIV infection, Hepatitis B and C infection [3].

Microscopically epithelial hyperplasia in FEH develops as focal acanthosis with thickening and elongation of the rete ridges. The surface keratinocytes have mitosoid bodies and koilocytic alterations. The ridges are confluent, broader, and occasionally formed like a club. Throughout the epithelium, there are areas where the cells show marked vacuolization (koilocytosis). This is most pronounced in the upper portion of the epithelium, but it may extend into the broadened rete ridges [8]. Similar findings were noted in our patients.

The current gold standard for HPV identification is molecular detection of HPV DNA. DNA polymerase amplifies the viral DNA in vitro to produce a large amount of target, which is thereafter directly seen on the gel and sequenced to identify the genotype [9-10].

In our case, HPV DNA detection and PCR sequencing of the tissue sample was done which was positive for HPV DNA

and showed HPV 84 genotype. To the best of our knowledge, this is the first case of Heck's disease caused by HPV 84.

As FEH is a benign condition, it resolves spontaneously in majority of cases. In persistent and severe cases, several treatment modalities like cryotherapy, electrocoagulation, treatment with carbon dioxide laser, topical treatment of retinoic acid and systemic treatment with interferon- α are useful [11].

Conclusion

This case report reveals a novel association between HPV 84 and FEH presenting in an adult. As a dermatologist, we need to broaden the differentials for raised lesions over the buccal mucosa to include FEH. Histopathology and PCR for HPV DNA with HPV typing are essential to diagnose Heck's disease.

Disclosure Statements

The authors hereby declare no Conflict of Interest.

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