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The spectrum of focal epithelial hyperplasia—a report of two cases

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Abstract

Focal epithelial hyperplasia is a rare, benign, and asymptomatic disorder, characterized by soft papules on the oral cavity. It is primarily associated with human papillomavirus genotypes 13 and 32. It most commonly affects children and young adults. When it affects young adults, it is important to differentiate it from oral *condyloma acuminata*. Its diagnosis may be made clinically, but histologic examination and PCR genotyping are often useful. Treatment is not always mandatory.

Keywords: focal epithelial hyperplasia, human papillomavirus, Heck disease

Introduction

Focal epithelial hyperplasia (FEH), also termed Heck disease, is a rare, benign, and asymptomatic disorder, characterized by soft, sessile, white-to-pinkish papules on the oral cavity [1]. It occurs most frequently on the labial and buccal mucosa, lower lip, tongue, and less often on the upper lip, gingiva, and palate [2]. It is primarily associated with human papillomavirus (HPV) genotypes 13 and 32 [3]. Risk factors for this condition include poor oral hygiene, low socioeconomic status, nutritional deficiency, and immunodeficiency [1]. It most commonly affects children and young adults [4]. There is no difference between the sexes. Some reports suggest familial clustering of cases [3]. Prevalence of this disease varies according to the geographic region. It appears to be most common in individuals of American Indian or Inuit descent [5].

Case Synopsis

Our first patient was a 24-year-old, HIV-positive smoker who presented with a 3-month history of asymptomatic soft, whitish, flat-topped papules bilaterally on the buccal mucosa (**Figure 1**). He was undergoing highly active antiretroviral therapy, with sustained virologic response. Histopathology showed acanthosis, hyperkeratosis, papillomatosis, mitosoid bodies, and viral cytopathic effects (**Figure 2**). Polymerase chain reaction (PCR) confirmed the presence of an HPV genotype not associated with oral condyloma acuminata (CA). Cryosurgery was proposed.

Our second patient, a 26-year-old man, with no smoking habits, presented with a 2-month history of

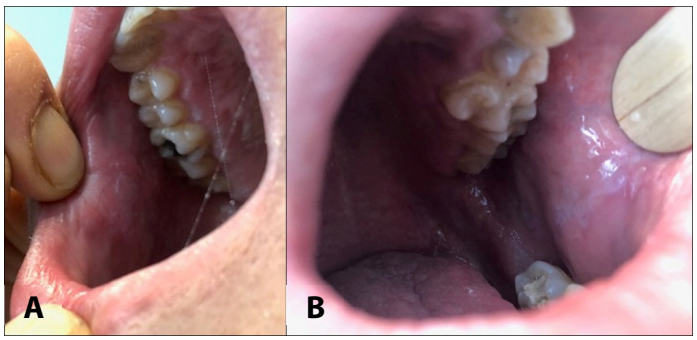

Figure 1. *Soft, whitish, flat-topped papules bilaterally on the buccal mucosa.*

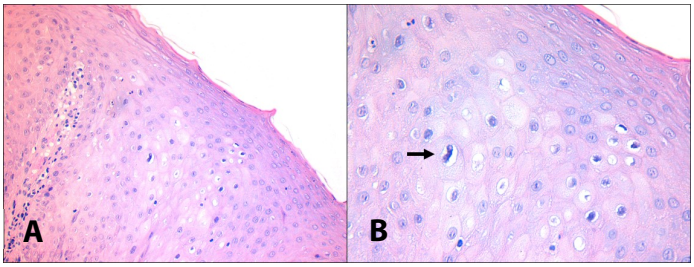

Figure 2. *H&E histopathology.* ***A)*** *Stratified squamous epithelium with papillomatosis, 100×.* ***B)*** *Mitosoid bodies (arrow), 200×.*

asymptomatic smooth, sessile white papules on the tip of the tongue (**Figure 3**). Medical history and physical examination were otherwise unremarkable. Histopathology displayed viral cytopathic effects, mitosoid bodies, and low-grade dysplasia (**Figure 4**). Polymerase chain reaction failed to detect HPV DNA in the tissue. The diagnosis of FEH was established based on clinical and histopathological findings. The patient was successfully treated with cryosurgery, with no relapse during a 6-month follow-up.

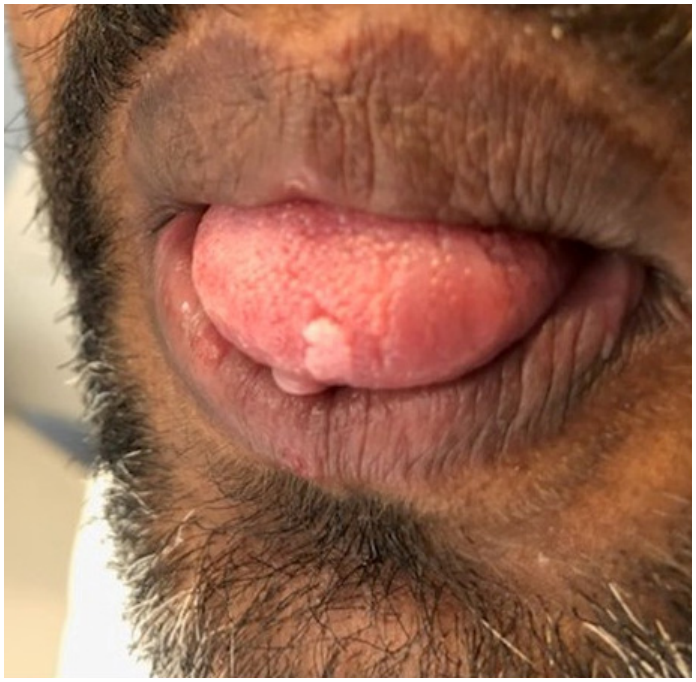

Figure 3. *Smooth, sessile white papules on the tip of the tongue.*

Case Discussion

A diagnosis of FEH may be made clinically, but histologic examination and PCR genotyping are often useful, mainly to differentiate it from oral CA, which contrary to FEH, are sexually transmitted and may be associated with a higher risk of malignancy. Furthermore, Heck disease only occurs in the oral mucosa. Histologically, the differences may be subtle, but elongated rete ridges with mitosoid bodies (keratinocytes with dense pyknotic nuclei or with a peripheral nuclear clearing with fragmented chromatin, which at low power can appear similar to a mitosis) are characteristic of FEH [2]. Nevertheless, distinctive histopathologic findings may not be represented in the biopsy sample and PCR may fail to detect HPV DNA, making the diagnosis a challenge. In over 90% of cases, HPV-13 or -32 is detected [6]. However, a diagnosis of Heck disease can be made in the absence of HPV detection, based on clinical and histopathological findings.

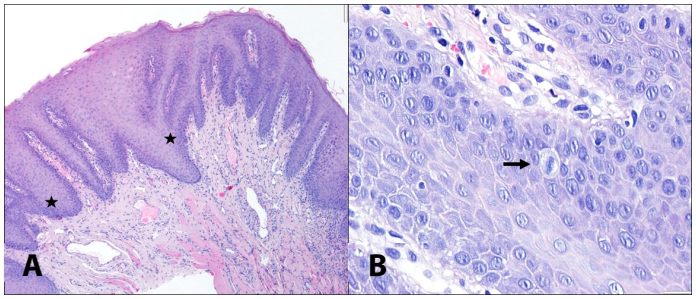

Figure 4. *H&E histopathology.* ***A)*** *Stratified squamous epithelium with broad anastomosing rete ridges (asterisks), focal keratinization and papillomatosis, 40×.* ***B)*** *Mitosoid bodies (arrow), 200×.*

Focal epithelial hyperplasia usually lasts for several months, sometimes years. Multiple therapeutic options are available, including cryosurgery [2]. However, treatment is not always mandatory, since this is a benign and asymptomatic condition.

Conclusion

Focal epithelial hyperplasia is a rare, benign, and asymptomatic disorder which should be part of the differential diagnosis of papules of the oral cavity.

Potential conflicts of interest

The authors declare no conflicts of interest.

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