Case Report

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Focal Epithelial Hyperplasia (Heck Disease) Associated with AIDS

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Key Words

Focal epithelial hyperplasia
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HIV infection

Abstract

Focal epithelial hyperplasia (FEH) of the oral mucosa occurring in a [HIV-infected](#) man is described. Molecular biology disclosed an HPV-32 type in oral lesions. The association of FEH and AIDS is uncommon although many HPV subtypes may manifest during HIV infection.

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Focal epithelial hyperplasia (FEH) of the oral mucosa was defined as circumscribed slightly elevated pink or white papules located mainly on the lower lip and buccal mucosa [1]. Most commonly two types of human papillomavirus, HPV-13 and HPV-32, were recognized in FEH and considered as the causative agents [2]. We report a new case of an HPV-32 infection correlated with oral FEH developing during HIV-infection.

Case Report

A 37-year-old Caucasian homosexual man with known AIDS since 1987 presented with flat smooth oral papules on the mucosal part of the upper and lower lip. The tongue remained unaffected and the general examination disclosed only discrete papilloma on the chin. At the time of examination biological evaluation of the immune status revealed: CD4 lymphocytes 72/mm³; CD4/CD8 ratio 1/7; positive P24 antigen 225 pg/ml.

A general treatment with zidovudine and sulfamethoxazole-trimethoprim was followed for 6 months.

We first considered the mucosal lesions as atypical oral hairy leukoplakia and therapy with aciclovir (1 g/day) was begun. A month

Fig. 1. Lesions of FEH on the lower labial mucosa.

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later, the mucosal papules were unaffected and the treatment was stopped.

Biopsy of oral papules was made because of new lesions on the mucosal surface of the cheek. Routine histological examination revealed epidermal hyperplasia with acanthosis and papillomatosis. A few cells were dyskeratotic, some with a clear cytoplasm and an irregular voluminous nucleus and marginated chromatin. All these changes were compatible with a viral origin.

In situ hybridization was also performed as previously described [3]. Epstein-Barr virus (EBV) genome was detected with bio BamHI W cDNA probe.

A strong signal with FITC-labeled anti-sense BHLF1 oligonucleotide probes demonstrated a lytic phase of EBV infection; labeling with EBER 1/2 oligoprobe remained negative and was supportive of absence of an EBV latent cycle.

Molecular cloning of extracted DNA found an HPV-32 band pattern (G. Orth, Institut Pasteur, Paris, France).

During the 3 following years, no further specific therapy was initiated and the lesions spontaneously resolved, but recurrences were noted.

Histological and virological examinations were repeated during this period on the newly developed lesions, showing similar results concerning morphological and virological analysis, but EBV remained undetectable.

Discussion

FEH of the oral mucosa was first described in 1965 by Archard et al. [1]. The diagnosis is based on clinical features, histology and detection of HPV types 13 and 32. However, additional HPV types have been detected in FEH (types 1,6-related, 11, 16, 18) [4, 5] and the clinical picture may be almost indistinguishable from condylomata of the oral cavity [6].

FEH occurs mostly among American Indians and Eskimos of Greenland but is sporadically reported in Caucasians [4]. An inherited predisposition is further supported by cases of familial recurrence with similar HPV types [7].

Initial reports of FEH postulated an associated undefined immunodeficiency [1]. In some cases, FEH has developed in immune-deficient patient: in 1 case [8], FEH occurred after immunosuppressive treatment, the other [9] developed in 2 siblings with leukocyte adhesion deficiency.

Unusual HPV-associated oral lesions have been investigated in HIV-positive patients [10, 11] but to our best knowledge, only 1 case of FEH associated with HIV infection has previously been reported in the studies by Vilmer et al. [12].

In HIV infection, oral warts harbor different HPV types: if HPV-7 appears more prevalent in the studies by Greenspan et al. [10] and de Villiers [13], other HPV types, including 13, 18 and 32, were also detected without clinical, histomorphological and virological correlations. In our case the clinical appearance, the histological features and the natural course are highly suggestive of FEH. The virological analysis of two different biopsy specimens disclosed HPV-32. This HPV type is closely associated with FEH but also found in limited cases of condylomata acuminata and squamous cell papilloma [2, 14]. The simultaneous occurrence of EBV infection initially noted may be coincidental because of the high prevalence of both HPV and EBV infections during AIDS. A conspicuous clinical and virological assessment of mucosal lesions of HIV patients might provide further argument about the relationship between FEH and immunodeficiency.

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