Oral Hairy Leukoplakia in Healthy, Immunocompetent Individuals

Abstract:

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Abstract

Oral hairy leukoplakia (OHL), while typically associated with HIV infection and immunosuppression, is rarely seen in HIV negative immunocompetent individuals. We report on two cases of OHL in immunocompetent patients.

Introduction

OHL, first described in 1981 and caused by the Epstein Barr Virus (EBV), presents as white, shaggy, non-removable plaques, typically on the lateral borders of the tongue[5]. It was initially considered pathognomic of HIV infection and latterly of immunosuppression. However more recently it is increasingly being reported as a complication of localised immunosuppression with topical or inhaled steroids[7,9]. Rare cases have also been documented in HIV negative individuals with no underlying history of topical or systemic immunosuppression[3,4,8,9]. It has been postulated that age may be a factor in some of these cases and chronic reactivation of EBV has been found to be related to aging[10].

Case Reports

Patient 1 is a 70 year old male, married with three children, referred for assessment of a white area on the lateral border of the tongue present for six weeks. Relevant medical history included osteoarthritis, hypertension and hyperlipidemia treated with olmesartan, felodipine and pravastatin. There was no history of immunosuppressive therapy. He was a non-smoker but consumed 30-40 units of alcohol per week. He was edentulous, with full upper and lower dentures. Denture hygiene was good. Examination revealed a soft, white, 4mm by 4mm plaque, on the right ventral surface of the tongue. An excisional biopsy was carried out and histopathological examination revealed parakeratosis with hyperplasia, candidal proliferation in the keratotic layer without any inflammatory reaction and groups of large cells with nuclear haloes suggestive of a viral effect. In-situ-hybridisation (ISH) identified EBV and a diagnosis of OHL was made. Investigations were carried out to identify any possible underlying immunodeficiency which could predispose to OHL including full blood count, electrophoresis, T-cell subsets, C-reactive protein, immunoglobulin levels and serology for HIV. The only finding was a decreased lymphocyte count of 1.1 ×109/l (normal range 1.5 â 3.5 ×109/l). HIV serology was negative initially and at retest six months later. There has been no recurrence post excision, the patient remains well and no underlying cause has been found.

Patient 2 is a 55 year old male, married with one daughter, referred for assessment of a white patch on the left lateral border of the tongue. Relevant medical history included well-controlled asthma for which he used a beclomethasone dipropionate inhaler twice daily. He was a non-smoker and did not consume any alcohol. Examination revealed a raised, creamy-white, soft, 50mm by 20mm plaque on the left lateral border of the tongue. There was also median rhomboid glossitis, denture stomatitis under an upper partial denture, and erythematous candidiasis of the dorsum of the tongue.

An incisional biopsy of the white plaque was carried out, swabs taken of the tongue and palate and instructions on inhaler technique and denture hygiene given. Histology showed hyperplasia with sheets of ballooned epithelial cells, parakeratosis and no inflammation. ISH was strongly positive for EBV in the superficial epithelial cells giving a diagnosis of OHL. Investigations to identify any possible systemic immunodeficiency were then carried out, as in the previous case. These revealed a raised white cell count of 13.0 ×109/l (normal range 4.0 â 11.0 ×109/l) with neutrophilia of 10.2 ×109/l (normal range 2.0 - 7.5) and raised immunoglobulin E of 198.0 u/ml (normal range 0.0 â 100.0 u/ml). HIV testing was negative initially and on retest six months later. Swabs grew significant levels of Candida albicans which was treated with nystatin suspension 100,000 units/ml four times daily for two weeks. The OHL was treated successfully with oral valciclovir. At most recent review there has been no recurrence, no underlying predisposing factor has been found and the patient remains well.

Discussion

While initially considered pathognomic of HIV infection and later of systemic immunosuppression, OHL is increasingly being reported in HIV-negative individuals. Some of these may arise as a complication of localised immunosuppression due to the use of topical or inhaled steroids, as in our second case. Chronic EBV reactivation related to aging may be a contributing factor in those without any identifiable underlying cause[10]. Histological findings in OHL are non-specific and include epithelial hyperplasia, acanthosis, hyperparakeratosis and koilocyte-like sub-corneal cells[1]. Definitive diagnosis requires demonstration of EBV in the lesional epithelium[2,6] using ISH or polymerase chain reaction (PCR) technology. OHL remains an important condition to recognise and, while it can occur in immunocompetent individuals, should always prompt a search for an underlying cause. It is possible that increased recognition may lead to further cases in immunocompetent individuals being reported.

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