Hairy leukoplakia in a child with AIDS—a rare symptom: case report

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Introduction

Hairy leukoplakia (HL) rarely has been described in the HIV+ pediatric population. This report details the presentation of HL in an otherwise healthy P2ADF AIDS 4-year-old male. The course of HL in this patient is described.

Case report

This 4-year-old Caucasian male developed normally until age 4 months, at which time he was diagnosed with pneumocystis carinii pneumonia, which was successfully treated with Bactrim, (Roche). By the end of his first year, he had suffered from pneumococcal bacteremia, septic shock, intermittent hypertension, hyperkalemia, and adrenal insufficiency. Zidovudine (AZT™—Burroughs-Wellcome, Research Triangle Park, NC) was added to the medication program. Within the following months, he developed complications to Bactrim prophylaxis, which then was replaced by intravenous Pentamidine (Fujisawa, Deerfield, IL). At 1 1/2 years old, he suffered a cardiac arrest secondary to hyperkalemia, later developed cardiomyopathy and was placed on Digoxin (Burroughs Wellcome, Research Triangle Park, NC). At age 2 years, a renal biopsy was performed to determine the etiology of his continued hypertension. Subsequent to the biopsy, he developed renal bleeding requiring a right nephrectomy. At age 2 1/2, the AZT was replaced by Dideoxyinmodine (DDI™—Bristol Myers, Evansville, IN) due to AZT toxicity.

At age 3 1/2 (December 1991), his mother noted multiple white patches on his tongue. Subsequently, he was examined by the Pediatric Infectious Disease Clinic. The physical evaluation was unremarkable except for bilateral white coating of his tongue, minimal anterior cervical adenopathy, and mild bilateral parotid gland enlargement. He was placed on a therapeutic trial of Ketoconazole (Janssen Pharmaceuticals, Titusville, NJ) after cultures of the lesions proved to be candida (OC). Laboratory investigations were unremarkable except for T-cell results of CD4:977 and CD8:787 respectively (ratio of 1.241).

In February 1992, he was referred to the Pediatric Dental Clinic for evaluation of residual tongue lesions, which the mother described as different from the candidiasis. He was a well-nourished child, with the appearance of the head and neck similar to that of the previous medical examination. Lips, buccal mucosa, pharynx, and extraoral soft tissues appeared normal except for the tongue. He had excellent oral hygiene, normal gingival health, and a complete noncarious primary dentition in good occlusion. The tongue lesions appeared as uneven, noncircumscribed, slightly elevated pale white patches that extended over the dorsum and bilaterally along the lateral borders (Fig 1, 2). They were not inflamed and resisted removal with gauze or toothbrush. The parent reported no discomfort with eating or speech. T-cell results were CD 4: 1421 and CD 8: 1624 (ratio of .875).

Smears from the lesions were placed on silare-coated
glass slides, (ONCOR, Gaithersburg, MD), air dried, and then fixed in 10% neutral buffered formalin (10% formalin in 0.1M sodium phosphate buffer, pH 7.0), for 15 hr. Protease digestion was done after fixation, using 2 mg/ml pepsin (Life Technologies, Gaithersburg, MD) digestion in 0.01N HCl at room temperature for 12 min. Epstein-Barr virus (EBV)-DNA analysis was done using polymerase chain reaction in situ hybridization, as previously reported.\textsuperscript{2} Examination of the cytology with routine staining revealed many squamous cells, some with slightly enlarged nuclei, scattered lymphocytes and rare neutrophils. EBV DNA was detected in squamous cells and not in lymphocytes. Detection of EBV DNA is consistent with the diagnosis of oral hairy leukoplakia.

In May 1992, the patient was admitted to University Hospital with a complaint of fever for three days and was diagnosed with pneumococcal sepsis. He responded to a 14-day course of antibiotic treatment and was discharged. At admission, his tongue lesions had partially regressed and no candidiasis was noted. T-cell results were CD4:1245 and CD8:1117 (ratio of 1.115). The HL has continued to resolve and as of December 1992, no traces of the lesions had been observed. He remains in stable health.

**Discussion**

OC is observed in approximately 15–40% of all children with HIV infection.\textsuperscript{3,4} The clinical manifestations are variable, ranging from small discrete patches to a diffuse pseudomembrane involving all of the oral mucosa. A more chronic form can develop on the dorsum of the tongue leading to loss of papillae. OC must be differentiated from HL.\textsuperscript{7} Most frequently, HL ranges from white, finely filamentous to papillary lesions along the lateral margins of the tongue to lesions that may extend to cover almost the dorsum. In adults, these lesions have been observed along the buccal mucosa, floor of the mouth, and palate. However, the lateral borders of the tongue are the most frequent sites.\textsuperscript{8,9} HL may regress during a course of antiretroviral therapy and/or concurrent improvement in cellular immunity.

Similar to the pediatric case previously reported by Greenspan et al., this child with AIDS experienced spontaneous remission of the HL after resolution of the OC with antifungal medication. In our patient, only after his candidiasis was treated were the residual lesions (HL) able to be identified. PCR of the lesions were positive for EBV, a well-known marker for hairy leukoplakia.\textsuperscript{8,9} While HL is felt to be a predictor of progression of HIV infection to AIDS in adults,\textsuperscript{7,8} this has not been observed in the pediatric population. When our patient’s transiently inverted CD cell ratio (and numbers) improved, the HL lesions resolved. We feel that HL may be more common than reported, but may be difficult to confirm due to overlying candidiasis. In addition, HL may appear, only transiently, secondary to intermittent changes in CD4 cell numbers.

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