

## An Uncommon Focal Epithelial Hyperplasia Manifestation

**Lourdes dos Santos-Pinto, DDS, PhD**    **Elisa Maria Aparecida Giro, DDS, PhD**  
**Cyneu Aguiar Pansani, DDS, PhD**    **Junia Ferrari, DDS, MS**  
**Elaine Maria Sgavioli Massucato, DDS, PhD**  
**Luis Carlos Spolidório, DDS, PhD**

### ABSTRACT

Focal epithelial hyperplasia is a rare, contagious disease associated with infection of the oral mucosa by human papillomavirus types 13 or 32, characterized by multiple soft papules of the same color as the adjacent normal mucosa. It mainly affects the lower lip, buccal mucosa, and tongue. The purpose of this case report was to describe a rare verrucal lesion located in the upper gingiva that was clinically and histologically consistent with focal epithelial hyperplasia.

(J Dent Child 2009;76:233-6)

Received August 31, 2008; Last Revision November 6, 2008; Revision Accepted November 7, 2008.

KEYWORDS: ORAL PATHOLOGY, FOCAL EPITHELIAL HYPERPLASIA, HUMAN PAPILLOMAVIRUS

**F**ocal epithelial hyperplasia (FEH) or Heck's disease is a rare infection of the oral mucosa associated with human papillomavirus (HPV) types 13 and 32, which mainly affects children and young adults.<sup>1,2</sup>

The first reported disease in the mouth described multiple nodular elevations in the oral mucosa observed in Navajo Indians. These lesions were similar to those described in South American Indians and Eskimos of Greenland and Alaska.<sup>3,4</sup>

Clinically, the FEH is characterized by multiple painless, well-defined, soft papules of the same color as the adjacent normal mucosa affecting mainly the lower lip, buccal mucosa, and tongue, and less often the upper lip, gingiva, and palate.<sup>5-9</sup>

Diagnosing this condition may be difficult due to a lack of clinical symptomatology and historical details.

The histological assessment becomes important and allows the observation of lesion characteristics, such as hyperparakeratosis, acanthosis, elongation and anastomosis of the rete ridges, classic koilocytes, perinuclear cytoplasmic halos, and nuclear dysplasia.<sup>4,10</sup>

The purpose of this case report was to describe an unusual case of focal epithelial hyperplasia in a young Brazilian child with generalized involvement of the upper gingiva.

### CASE REPORT

Clinical examination of a 3-year old boy revealed a localized mucosa enlargement from the primary maxillary right lateral incisor's distal surface to the primary maxillary left central incisor's distal surface in the maxillary arch. The lesions presented flat-topped nodules with a finely stippled and slightly verrucal surface (Figure 1). The X ray examination showed no bone alteration in the lesion region.

The patient's mother reported that lesions had been present for several months, were painless, and had rapidly increased in size, presenting a "cauliflower" appearance.

The child's medical history revealed the presence of anemia and the use of multivitamin supplements and

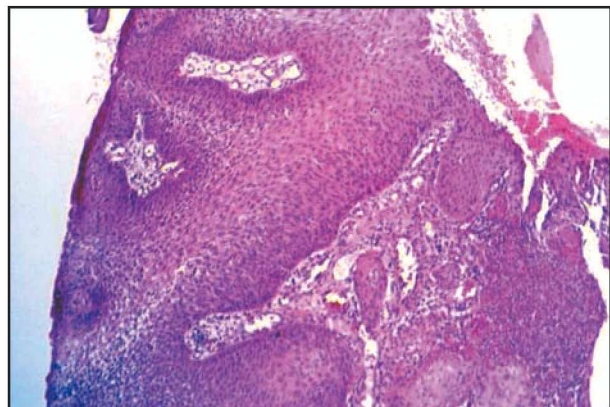
*Drs. Santos-Pinto, Giro, and Pansani are associate professors and Dr. Ferrari is a PhD student, all in the Department of Orthodontics and Pediatric Dentistry, Dr. Massucato is associate professor in the Department of Diagnosis and Surgery, Dr. Spolidório is associate professor in the Department of Physiology and Pathology, all at Araraquara Dental School, University of São Paulo State, São Paulo, Brazil. Correspond at Dr. Santos-Pinto at [lpinto@foar.unesp.br](mailto:lpinto@foar.unesp.br)*



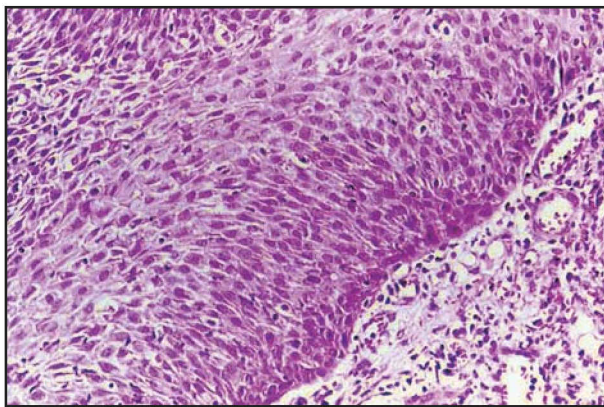
**Figure 1.** Lesions in the upper gingiva.



**Figure 2.** Clinical aspect of the lesion after 15 days.



**Figure 3.** Histologic section showing epithelial hyperplasia.



**Figure 4.** Presence of acantosis and mitosoidal cells.



**Figure 5.** Remission of the lesions after treatment.



**Figure 6.** 24 months follow-up.

ferric sulfate. The child and his father presented traces of sickle cell disease.

The clinical initial impression was that this was a peripheral giant cell lesion; it was also thought to be papilloma and HPV lesions. The mother's medical history revealed that she was HIV- and HPV-negative, and the child tested negative for both conditions. The lesion treatment was immediately instituted and consisted of the professional dental prophylaxis, maintenance of oral hygiene, and topical application of chlorhexidine gluconate (0.12%) once a day.

The patient returned after 15 days, when it was observed that the lesions had grown more in volume than in extension involving the lateral aspect of the primary

maxillary right lateral incisor (Figure 2). Once anemia was treated and the patient's systemic conditions were stabilized, incisional biopsy was performed in the right lateral incisor's distal papilla. Removing the lesion completely was not possible because of its extension and the child behavior.

The histological exam showed acanthosis of the oral epithelium and confluence. Areas with coilocytas cells and cells showing a mitosis-like nuclear degeneration (mitosoidal cells) were observed. The basal layer was undamaged, and the connected tissue presented chronic inflammatory infiltration. The histological characteristics determined the diagnosis of FEH (Figures 3 and 4).



Five weeks after beginning treatment, complete remission of the lesions was observed. At this appointment, the mother reported an important fact to elucidate the etiology of this unusual disease. Two months before the lesions' appearance, the child was seen blowing up a used condom found in the street.

The use of chlorhexidine was continued for 15 more days, and the lesions were totally repaired (Figure 5). This treatment provoked tooth stains, which were removed with ultrasonic scalers and dental prophylaxis using a rubber cup and prophylactic paste at low speed.

The patient was followed for 2 years, and recurrence of the disease was not observed (Figure 6).

## DISCUSSION

HPV is an acronym used to identify the human papillomavirus, which infects the skin and mucosas and might induce the formation of both benign and malignant tumors. The infection starts when the virus penetrates the host through microinjuries.<sup>4</sup>

The FEH is a benign infectious disease caused by HPV, and its manifestation is exclusively in the mouth.<sup>5</sup> The correct diagnosis must exclude other benign oral and oropharynx lesions associated with HPV, such as squamous cells papilloma, condiloma acuminata, common warts, and malignant oral lesions, especially squamous cell carcinoma.<sup>4</sup>

The diagnosis is based on clinical and cytological characteristics and also on molecular biology exams, such as immunohistochemistry, in situ hybridization, Southern blot, and PCR (Polymerase chain reaction), which detect the presence of HPV types 13 and 32 in the lesions.<sup>11</sup>

In the present case report, the lesion was located in the upper gingiva, which is uncommon. The lesions are commonly frequent in the lower lip, buccal mucosa, upper lip mucosa, and tongue.<sup>5-7</sup> The lesion presented a slightly verrucal surface, while the most frequent appearance is as soft papules of the same color as the adjacent normal mucosa.<sup>4-8</sup>

Although FEH etiology is related to a virus, it has been suggested that genetic predisposition can be involved, since there is familiar occurrence of a predilection in some ethnic groups.<sup>6,10,12</sup> Concerning the reported case, we cannot confirm the genetic association because there was no other case observed among the family and the family history did not reveal Indian descent.

Other risk factors associated with viral infection were evaluated and indicated toothbrush and spoon sharing.<sup>2</sup>

FEH is only rarely associated with immunodeficiency or immunosuppression.<sup>13</sup> The immune status of our patient was normal. In sickle cell disease, however, the cells' short life span often results in anemia and the red blood cells cause blockages and obstruct the blood circulation. This causes vascular occlusion, tissue lesions, and infection problems.<sup>14</sup> Since blood vessels are spread

all over the body, there is a risk of lesions everywhere, underlining the importance of early diagnosis of FEH.

The treatment consisted only of oral hygiene and topical application of chlorhexidine due to the patient's age and the possibility of spontaneous lesion remission. Chlorhexidine was prescribed to control plaque accumulation. The diminished effect of chlorhexidine on structured biofilm reinforced the necessity of biofilm disruption before the initiation of chlorhexidine mouth-rinse.<sup>15</sup> The risk of tooth staining from chlorhexidine was managed by regularly using ultrasonic scalers and polishing with rubber cups.

Surgical removal is indicated for esthetic purposes and when lesions are frequently traumatized or interfere with function.<sup>12,16</sup> Other treatment options are cryotherapy, electrocoagulation, laser, chemical agents such as retinoic acid, and immunostimulants such as interferons.<sup>16-18</sup>

It is important to verify whether the viral infection was sexually transmitted, as this could indicate sexual abuse. In the present report, the infection was caused by a contaminated condom inappropriately discarded in the street and found by the child.

The main histological characteristics include epidermal acanthosis and parakeratosis, thickening and elongation of the rete ridges, and pronounced koilocytosis with characteristic mitosoid figures. These lesions may persist for several years, but do not become malignant and tend to regress spontaneously.<sup>2</sup>

Recurrence is unpredictable, and the continued follow-up of patients is essential. When relapse occurs, it is not clear whether a new infection is present or whether it is due to viral latency or the reduction of the patient's immune response.<sup>4</sup> The patient described here has been followed for a period of 24 months without any recurrence.

## REFERENCES

1. Tying, SK. Human papillomavirus infections: Epidemiology, pathogenesis, and host immune response. *J Am Acad Dermatol* 2000;43:518-26.
2. Cuberos V, Perez J, Lopez CJ, et al. Molecular and serological evidence of the epidemiological association of HPV 13 with focal epithelial hyperplasia: A case-control study. *J Clin Virol* 2006;37:21-6.
3. Archad HO, Heck JW, Stanley HR. Focal epithelial hyperplasia: An unusual mucosal lesion found in Indian children. *Oral Surg* 1965;20:201-12.
4. Castro T P, Bussoloti Filho I. Prevalence of human papillomavirus (HPV) in oral cavity and oropharynx. *Rev Bras Otorrinolaringol* 2006;72:272-82.
5. Morrow DJ, Sandhu HS, Daley, TD. Focal epithelial hyperplasia (Heck's disease) with generalized lesions of the gingiva. A case report. *J Periodontol* 1993; 64:63-5.

6. Ledesma-Montes C, Vega-Memije E, Garcés-Ortíz M, Cardiel-Nieves M, Juárez-Luna C. Multifocal epithelial hyperplasia: Report of nine cases. *Med Oral Patol Oral Cir Bucal* 2005;10:394-401.
7. Borborema-Santos CM, Castro MM, Santos PJB, Talhari S, Astolfi-Filho S. Oral focal epithelial hyperplasia: Report of five cases. *Braz Dent J* 2006; 17:79-82.
8. Terezhalmay GT, Riley CK, Moore WS. Focal epithelial hyperplasia (Heck's disease). *Quintessence Int* 2001;32:664-5.
9. Jablonska S, Majewski S. Demonstration of HPV in long-standing Heck's disease with malignant transformation. *Eur J Dermatol* 2000;10:235-6.
10. González LV, Gaviria AM, Sanclemente G, et al. Clinical, histopathological, and virological findings in patients with focal epithelial hyperplasia from Colombia. *Int J Dermatol* 2005;44:274-9.
11. Jayasooriya PR, Abeyratne S, Ranasinghe AW, Tilakaratne WM. Focal epithelial hyperplasia (Heck's disease): Report of two cases with PCR detection of human papillomavirus DNA. *Oral Dis* 2004; 10:240-3.
12. Martins WD, Lima AA, Vieira S. Focal epithelial hyperplasia (Heck's disease): Report of a case in a girl of Brazilian Indian descent. *Int J Paediatr Dent* 2006;16:65-8.
13. Steinhoff M, Metze D, Stockfleth E, Luger TA. Successful topical treatment of focal epithelial hyperplasia (Heck's disease) with interferon- $\beta$ . *Br J Dermatol* 2001;144:1067-9.
14. Adams-Graves P, Ostric EJ, Martin M, Richardson P, Lewis JB Jr. Sickie cell hospital unit: A disease-specific model. *J Healthc Manag* 2008;53:305-17.
15. Zanatta FB, Antoniazzi RP, Rösing CK. The effect of 0.12% chlorhexidine gluconate rinsing on previously plaque-free and plaque-covered surfaces: A randomized, controlled, clinical trial. *J Periodontol* 2007;78:2127-34.
16. Flaitz, CM. Focal epithelial hyperplasia: A multifocal oral human papillomavirus infection. *Pediatr Dent* 2000;22:153-4.
17. Steinhoff M, Metze D, Stockfleth E, Luger T A. Successful topical treatment of focal epithelial hyperplasia (Heck's disease) with interferon-[beta]. *Br J Dermatol* 2001;144:1067-9.
18. Durso BC, Pinto JMV, Jorge Jr J, Almeida OP. Extensive focal epithelial hyperplasia: Case report. *J Can Dent Assoc* 2005;71:769-71.

Copyright of Journal of Dentistry for Children is the property of American Academy of Pediatric Dentistry and its content may not be copied or emailed to multiple sites or posted to a listserv without the copyright holder's express written permission. However, users may print, download, or email articles for individual use.