An Uncommon Focal Epithelial Hyperplasia Manifestation

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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.
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KEYWORDS: ORAL PATHOLOGY, FOCAL EPITHELIAL HYPERPLASIA, HUMAN PAPILLOMAVIRUS

Focal 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.[1,2]

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.[3,4]

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.[5-9]

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 coilocytes, perinuclear cytoplasmic halos, and nuclear dysplasia.[4,10]

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

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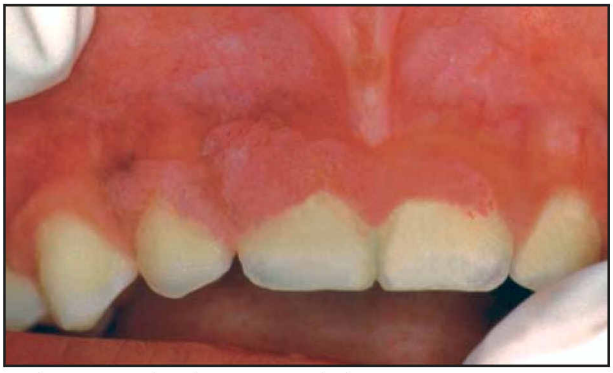

Figure 1. Lesions in the upper gingiva.

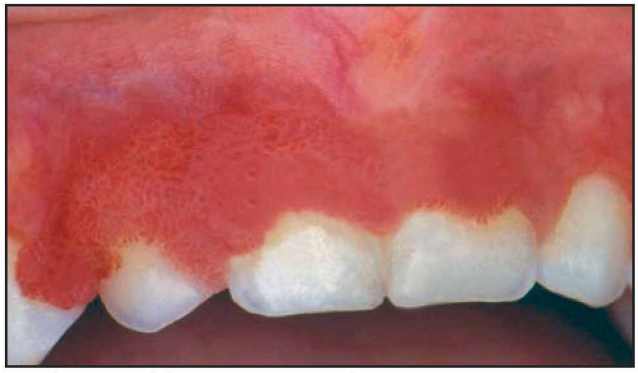

Figure 2. Clinical aspect of the lesion after 15 days.

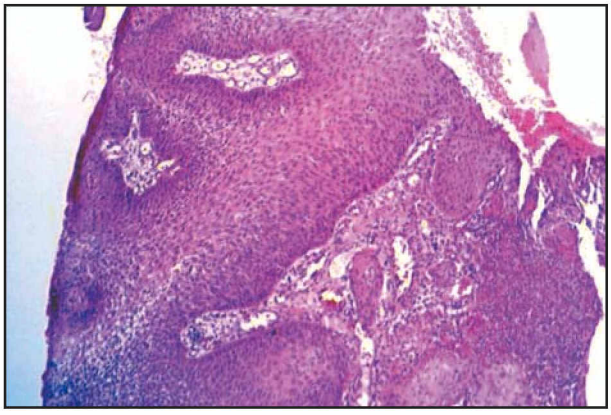

Figure 3. Histologic section showing epithelial hyperplasia.

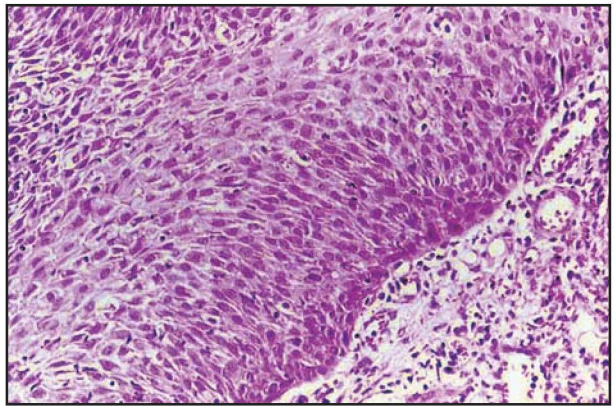

Figure 4. Presence of acantosis and mitosoidal cells.

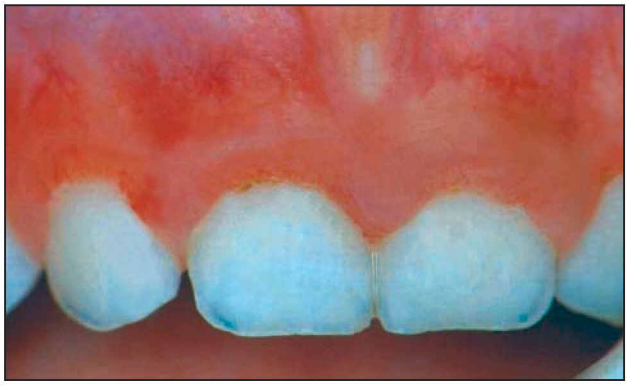

Figure 5. Remission of the lesions after treatment.

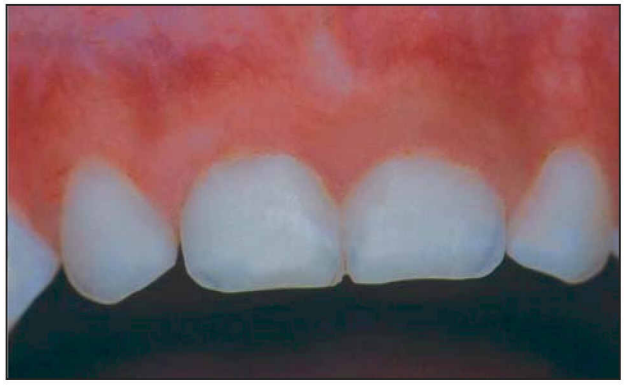

Figure 6. 24 months follow-up.

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 coilocitas 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 chlorexidine 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 papillo mavirus, 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.[4]

The FEH is a benign infectious disease caused by HPV, and its manifestation is exclusively in the mouth.[5] 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.[4]

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.[11]

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.[5-7] 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.[4-8]

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 ethic groups.[6,10,12] 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.[2]

FEH is only rarely associated with immunodeficiency or immunosuppression.[13] 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.[14] 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 chlorexidine 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 mouthrinse.[15] 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.[12,16] Other treatment options are cryotherapy, eletrocoagulation, laser, chemical agents such as retinoic acid, and immunostimulants such as interferons.[16-18]

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.[2]

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.[4] The patient described here has been followed for a period of 24 months without any recurrence.

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