Focal epithelial hyperplasia: A multifocal oral human papillomavirus infection

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Excluding gingival lesions, multifocal soft tissue enlargements in children are an unusual oral finding. Intraoral warts, irritation fibromas, and lesions associated with certain hamartoneoplastic syndromes may present as persistent and multiple nodules in this age group. The purpose of this case report is to illustrate an example of focal epithelial hyperplasia (FEH) and to discuss a reasonable differential diagnosis, based on the age of the patient, history and clinical features.

Case history
A 5 year-old Hispanic girl is referred for the evaluation of widespread mucosal changes that coincided with a serious car accident about a year ago. Clinical examination reveals coalescing plaques and flat-topped nodules with finely stippled to cobblestone and fissured surfaces. Although the lesions are distributed throughout the oral cavity, these mucosal enlargements are most prominent on the left buccal mucosa (Fig 1) dorsal tongue (Fig 2) and alveolar mucosa. This is the same side that had a previous history of an alveolar fracture with loss of the primary molars, persistent numbness and perioral scarring from the traumatic lacerations. A class III malocclusion with an openbite and aberrant tongue position (Fig 3) on the affected side, along with perioral grimacing are additional findings. These orofacial contortions and chronic sucking behaviors are suspected in the spread of these lesions. Except for some surface roughness, these soft nodules are asymptomatic but are increasing in size. The child’s medical history is remarkable for periodic headaches, stomach pain and behavioral problems that have been diagnosed since the car accident.

Clinical impression
Based on the clinical presentation and history, these mucosal changes are most consistent with oral warts, known as FEH or Heck’s disease. This site-specific disease represents a human papillomavirus (HPV) infection that is caused by HPV types 13 and 32.1 Usually a childhood condition, a predilection for Native Indians from North and South America, and Eskimos is observed in the United States.2 However it has been reported sporadically in many other populations and ethnic groups. A genetic predisposition and an immunocompromised host status, due to malnutrition and crowded living conditions, are thought to be contributing factors for this infectious disease.2,3 Although several reports demonstrate a familial tendency,2,4
which would favor an infectious disease transmission, larger epidemiologic studies fail to demonstrate this pattern. Clinically, FEH is characterized by multiple, nontender, papules, plaques and nodules that are soft to palpation and have a grainy, flat-topped surface. Although these enlargements are often multifocal and discrete, a cobblestone or fissured pattern is observed with coalescing sheets of oral warts. Bilateral involvement of the labial and buccal mucosa and dorsolateral tongue is the classic distribution, although any oral site may be affected. Repeated trauma of a cluster of lesions along the occlusal plane may produce a nodule with a papillary surface that is similar to a condyloma acuminatum.

Diagnosis

Incisional biopsy of a representative lesion, along with histopathologic evaluation, is recommended for the diagnosis of this persistent mucosal disease. In some cases when it is important to exclude a sexually transmitted disease, then special studies, such as DNA in situ hybridization for HPV types 13 and 32, should be performed on the surgical specimen.

Treatment

Management of these lesions should be conservative in children because spontaneous resolution may occur in a few months to several years. Surgical excision may be indicated for lesions that are frequently traumatized and/or for esthetic purposes. Other treatment modalities with variable effectiveness include cryotherapy, laser ablation, topical application of 25% podophyllin resin and vitamin therapy. Recurrence of these oral lesions, even following spontaneous regression, is common. It is uncertain whether these lesions recur due to latent infection, new infection or fluctuation of the immune response to this viral disease.

Differential diagnosis

In a child, it is often important to distinguish FEH from condyloma acuminata (CA) or venereal warts. This sexually transmitted wart is commonly found in the anogenital region and rarely occurs intraorally. Oral transmission of CA results from direct contact due to orogenital sex or self-inoculation from hand to mouth exposure. Another important route in a child includes perinatal transmission from infected mother to newborn. Of importance, the most common HPV types associated with oral CA are 6 and 11. Clinically these venereal warts present as multiple, asymptomatic nodules with cauliflower or mulberry surfaces, although early lesions may appear as flat-topped, stippled nodules. A more localized and clustered distribution is observed with CA, in contrast to FEH.

When the lesions of FEH are more discrete and isolated, the soft tissue nodules are very similar to an irritation fibroma (focal fibrous hyperplasia). Typically these reactive, soft tissue enlargements have a smooth, pale surface and are most prominent along the occlusal plane of the buccal mucosa or lateral border of the tongue. In contrast to FEH, these nodules gradually increase in size and then remain static. A source for the chronic irritation is readily apparent by history or clinical examination. Lastly, several hamartoneoplastic syndromes have oral manifestations that are characterized by multiple soft tissue enlargements. Multiple endocrine neoplasia syndrome, type III, neurofibromatosis, tuberous sclerosis, epidermal nevus syndrome and Cowden syndrome are important examples of entities with mucocutaneous involvement.

Pediatric significance

FEH is an uncommon childhood disease with widespread oral involvement that may mimic CA and several syndromes with head and neck manifestations. It is important for the pediatric dentist to recognize that these oral lesions are caused by a HPV infection but they do not represent a sexually transmitted disease. In this present case, a definitive diagnosis with viral typing is advisable because the child is experiencing nonspecific head and abdominal pain, in addition to behavioral problems. Although the emotional and physical trauma from a disfiguring car accident may be the inciting cause for these symptoms, it is essential to verify that these oral warts are not a manifestation of sexual abuse.

References