Multifocal Epithelial Hyperplasia. An Unusual Lesion.

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Abstract

Multifocal epithelial hyperplasia is a virus related disease affecting the oral mucosa in children and there are reports of this disease in several ethnic groups of the world. The aim of this study is to present the clinical and pathological findings encountered during the review of one 8 years old, Mestizo boy. We found several elevated, soft, smooth surfaced lesions in the buccal mucosa and lower and upper lips mucosa. Microscopically, a parakeratinized, acantotic epithelium was observed. Characteristically, mitosis-like cells were present. His family was in poverty and two brothers and one cousin presented similar lesions.

Introduction

In 1956, Estrada reported on the presence of multiple intraoral soft tissue lesions discovered in Colombian Caramanta Indians [1]. Later, he reported on the same lesions in Katio Indians of El Chocó [2]. Reyes communicated that this disease was present in Guatemalan population; he diagnosed these lesions as verrucae of the oral cavity [3]. Several years later, Soneira and Fonseca [4] reviewed 160 Venezuelan Indians, of them, 54 patients presented papulo-nodular lesions in their oral mucosa. They must be credited with the first complete description of this entity, since they described in full the clinical and microscopic features, gave details on the exfoliative cytology findings and their article is profusely illustrated with numerous clinical pictures and photomicrographs. They suggested this disease was of viral origin. In the European dermatological literature there is the Stern’s report [5], he presented several cases with similar lesions in the intraoral soft tissues verrucae. In his article, Praetorius-Clausen [6] mentioned that in 1894, Helms described lesions resembling this disease in Greenlandic Eskimos. Unaware of the previously published Latin American and European reports on this disease, Archard et al and Witkop and Niswander [7,8] published 19 cases in the English language literature. In the last years, different reports of lesions found in American Indians [9-11], European [12-14], Asiatic [15,16] and African patients [17-19] had been published. Lesions clinically and microscopically similar to this disease were found in two chimpanzees and one rabbit [20,21]. In 2005, Ledesma-Montes et al suggested the most adequate name for this disease this disease is multifocal epithelial hyperplasia (MEH) and with this name is widely known in Latin America [10]. MEH has preference for appearing in children. Clinically MEH is characterized by the presence of multiple lesions, but rarely isolated lesions can be found [11,13,19,22]. MEH lesions are papulo-nodular, elevated, asymptomatic and smooth surfaced swellings, when they are of verrucous or papillomatoid surface they can be confused with papillomas. Usually, they are round or oval, well defined nodules which present no changes with the surrounding mucosa, most of them are 0.1 to 0.5 cm in diameter, but lesions measuring several centimeters are not rare, they are formed by coalescence of several minor nodules. They are slow growing lesions and malignant transformation has not been observed [11,22]. It is characteristic that nodules disappear when they are stretched and when tension is liberated, they come into view [11], a feature that has been considered pathognomonic for this disease. Previous reports communicated that MEH is more common in females; the more frequently affected locations are labial, lingual and buccal mucosa. Some authors reported it is more frequent in children [11,22] but there are some communications in adults [15,23]. It is well known that MEH lesions are frequently associated to human papillomavirus HPV types 13 and 32 [24-28]. Some immunocytochemical and in situ hybridization studies demonstrated the presence of VPH-16, VPH-11 [29], an 11-related HPV [27], VPH-1 [30] and VPH-6 [31]. Other reported associated factors are: a recessive gene, galvanic electricity from dental amalgams, tobacco chewing and lack of vitamin K [4,7,12,15,32,33].

Case Report

An eight years old boy assisted to the Oral Diagnosis Clinic of the Facultad de Odontología, UNAM in México City. His main complaint was the presence of several slightly elevated, sessile, soft, smooth surfaced lesions with the same color of the surrounding mucosa. They were asymptomatic, slow growing lesions appearing since two years ago. Lesions were located in both, lower and upper lips mucosa (Figure 1) and buccal mucosa (Figure 2). Characteristically, when we stretched the lesions, they
Oral hygiene was good and economic status was considered as poverty since monthly income of the family was below $200 US dollars. Two brothers and one cousin presented identical lesions. One lesion was biopsied by local anesthesia and immersed in aqueous neutral formalin solution for fixation during 24 hours and routinely processed to obtain 5 µ thick, paraffin embedded, H and E stained slides.

Microscopic examination showed the presence of parakeratinized squamous stratified epithelium with marked acantosis and thick epithelial rete ridges (Figure 3). Numerous koilocytes were observed and some mitosis-like cells identified (Figure 4). Connective tissue was formed by fibroblasts, collagen bundles, some chronic inflammatory cells, capillaries and small nerves. Clinical and microscopic features corresponded with a final diagnosis of multifocal epithelial hyperplasia.

Discussions and Conclusion

In the past, MEH was known as focal epithelial hyperplasia. This disease is well-known by Latin-American dental and medical practitioners. As the name MEH indicates, the most prevalent clinical finding is the presence of multiple nodular lesions in the oral mucosa. It affects lips, buccal mucosa, tongue and commissures [11,22] and is more common in children. Another factor associated to this disease is poverty [11,22]. To date, according to the first report published by Estrada [1] who suggested that MEH was of viral origin, actual data support that the presence of the human papillomavirus types 13 and 32 identified within the lesional epithelium [24,25,30,31], families with several affected relatives and its preference by economically poor population with deficient medical care and limited access to the health system [10,11,22], support the theory that a viral infection is the etiological agent of MEH.

It is important to diagnose adequately this disease, since it is frequently confused with the “florid oral papillomatosis”, vulgar verrucae, papilloma, condiloma acuminatum or lesions related to child abuse. Treatment for MEH is commonly the surgical excision, but application of liquid nitrogen was also used [10,11]. Archard et al suggested administration of sulfamides and vitamin A [7]. There is a general agreement that MEH lesions should not be excised, since it is a self-limiting disease and because lesions will disappear with the age of the patient. Only should be excised those lesions found in traumatic areas or those lesions which are traumatized by the patient. Other suggested treatment was the use of local cryotherapy by means of liquid nitrogen applications [11,22]. In our experience, all the patients, including those children with one lesion at first examination, more lesions will appear in the oral soft tissues in the following weeks or months and if they are adults, clinical interrogation will demonstrate that at the beginning of the disease the patient presented multiple lesions. It is important to note that during the interrogatory, all adult patients recognized that they presented several lesions during childhood [11].

We consider that the name FEH should be discarded and the name proposed by Ledesma-Montes et al [11] as multifocal epithelial hyperplasia should be adopted, since for the experience, all the patients have two or more lesions during the course of the disease. The use of this name has the advantage to describe both the main clinical feature of the disease (the presence of multiple lesions) and the main microscopic finding (epithelial hyperplasia). We agree with the proposal to abandon the eponym Heck’s disease since the first well documented cases were published by Estrada [1,2] and Soneira and Fonseca [4] should be credited as the first authors to describe MEH in full.

Our study group [10,11] reported that the presence of familial MEH lesions could be related to common use of knives, forks and spoons among the family members. Our assumption is confirmed by the results of the González-Losa et al study, showing that HPV 13 and 32 is present in the saliva of more than 73% of their studied population [35].

References

7. Archard HO, Heck JW, Stanley HR. Focal epithelial hyperplasia: An unusual oral mucosal lesion found in
Illustrations

Illustration 1

Fig. 1. Numerous lesions in lower lip mucosa.

Illustration 2

Fig. 2. Elevated lesions present in buccal mucosa.
Illustration 3

Fig. 3. Photomicrograph showing the microscopic features of the multifocal epithelial hyperplasia. H and E. 100X.

Illustration 4

Fig 4. Photomicrograph showing the presence of mitosis-like cells. H and E. 400X
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