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Case Report

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Multifocal Epithelial Hyperplasia, a Rare Oral Infection in the Iranian Population: A Case Report

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Abstract

Multifocal epithelial hyperplasia (MEH) or Heck's disease is a rare, benign, non-common, self-limiting viral infection of the oral mucosa that is caused by human papillomavirus (HPV), especially subtype 13 or 32. The onset of the disease varies widely between different geographic regions and different races. A 45-year-old male patient was admitted to Department of Oral Medicine of Yazd Dental School for dental examination. Routine extra oral examination revealed numerous papules in the lips. The lesions were multiple, scattered papules that were interconnected in some areas and resembled an exophytic mass. Subsequently, the family history was positive and the clinical features conducted us to the diagnosis of the Focal Epithelial Hyperplasia. Since this disease is rare in the Iranian population and this case is the 19th case in Iran, timely diagnosis and prevention of its transmission had the great importance. Therefore, promotion of dentists' awareness towards diagnosis and appropriate referral to oral medicines specialists were recommended.

***Keywords*:** Focal Epithelial Hyperplasia, Human Papilloma Virus, Heck's Disease, Oral, Hyperplasia

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Introduction

Multifocal epithelial hyperplasia (MEH) is a benign, non-common, self-limiting viral disease which was first described and reported by Archard et al. (1) Most cases of the disease have been reported in children, but it was seen in the adults with immunological problems (2). In order to the appropriate diagnosis, clinical and histopathologic features consideration is recommended (2, 3). Its clinical features can be characterized by multiple pink smooth surface broad base papules with a distinct margin (4, 5). In the most histopathologic examinations, 13 and 32 subtypes of human papillomavirus (HPV) have been identified (2, 3). Histopathologic features of MEH are distinct and

indicate HPV infection, which includes clear acanthosis, koilocyte cells, and mitotic cells. Long papillae, dilated capillaries, and mild lymphohistiocytic infiltration are also found in connective tissue (5). Genetic factors are also likely to affect these lesions (1, 3). However, its etiology is unclear and needs further investigation. Lesions in this disease are mostly found in the oral mucosa, especially on the lips, buccal mucosa, and tongue, but it was also reported in the skin and genital mucosa (1). The lesions are more common in Eskimos, North and South America (2), and are relatively rare in Asian populations (5). MEH is very rare in the Iranian population, and in this study we report a case of this disease in a 45-year-old Iranian male in Yazd city.

Case Presentation

A 45-year-old male patient was admitted to Department of Oral Medicine of Dental Faculty of Yazd University for dental examination. Routine extra oral examination revealed numerous lesions in the lips that the patient was ignorant and not aware of them. The patient stated that he had these lesions since he remembered and was trying to keep his beard and mustache long to cover the lesions. The lesions were multiple, scattered papules that were interconnected in some areas and resembled an exophytic mass. The lesion has pink, smooth, intact surface with approximately 1×1.5 cm dimensional of largest mass and located on the upper lip commissure (Figure 1-A, 1-B, and 1-C). Since the family history was positive, the clinical features conducted us to the diagnosis of the Focal Epithelial Hyperplasia. An appointment was made to remove the other lesions with a cutter, but unfortunately the patient was not presented due to the lesions were asymptomatic.

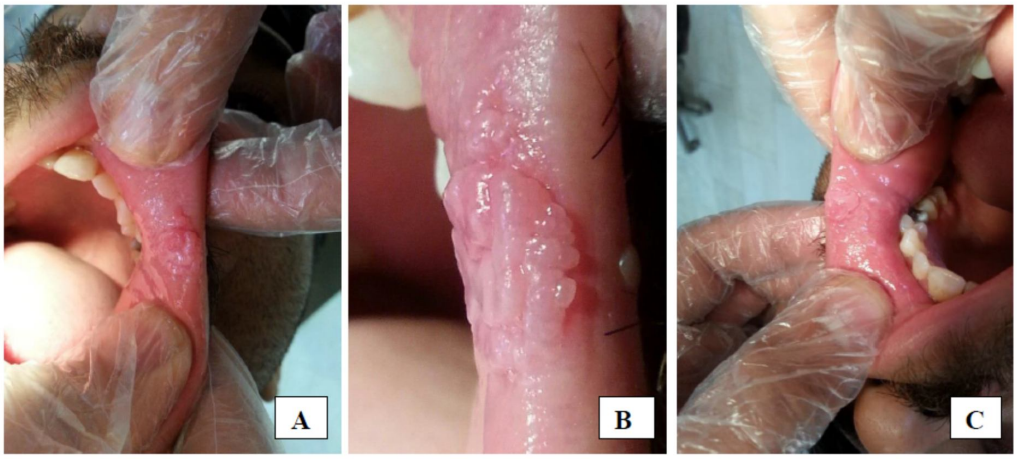

Fig. 1. Clinical features: A) Multiple and scattered papules were obvious. B) The lesions were interconnected in some areas and resembled an exophytic mass. C) The lesion has pink, smooth, intact surface located on the upper lip commissure

Discussion

Multifocal epithelial hyperplasia (MEH) is a rare viral disease with no identified specific etiology (1, 4); previously, etiologic factors such as geographical factors and nutrition were recommended (1). Human Papilloma Virus (HPV) as a possible etiologic factor has been identified due to clinical and histologic features of the lesions. Subsequent studies identified HPV 32 in the most non-keratinized oral lesions and HPV 13 in the most keratinized lesions (1, 5, 7-11). However, a case of this lesion has been reported in China which has no relation to HPV (5). Regards to its incidence among family members, genetic factors are also considered as other possible causes (1, 3, 12, and 13). Studies have

been carried out to investigate the prevalence of these lesions in different communities; according to the studies it was most prevalent in Eskimos (1). Recently, the susceptibility of organ transplant recipients to the development of MEH have been reported (12). In the Iranian populations, only two cases of MEH have been reported; one in a 12-year-old girl and another one in a 35-year-old man that both of them have multiple lesions on the lower lip and buccal mucosa (8). The present patient was a 45-year-old man as the third case of MEH in an Iranian family and 19th in Iran, who had MEH in his lower lip and commissural region. Although most of the cases have been reported in children, it was also seen in the adults with suppressed immune system (1, 3, and 13). Although some lesions are self-limiting, the course and duration of the disease are variable; some lesions may become larger, chronic, or even recur. So that further investigation is necessary in this situation (1, 12). Most of these lesions have been reported in the oral mucosa but it has been also reported in the other parts of the body such as the genital mucosa (1, 12). Furthermore, this lesion has been reported in animals (13). Various methods have been recommended to diagnose the MEH, including clinical and histopathological examination as well as molecular and PCR methods in order to detect HPV and its subtypes (1). The diagnosis of Focal epithelial hyperplasia (Heck disease) is very important because some diseases such as Inflammatory Fibrosis Hyperplasia, Inflammatory Papillary Hyperplasia, Verruciform Xanthoma, Verrucous Carcinoma, Cowden's disease, Condyloma Acuminatom, and Goltz-Gorlin syndrome are considered as the differential diagnosis of MEH (1, 6, 7). The first three lesions which are mentioned above are reactive lesions and in the most cases, they had a stimulating factor. Verrucous carcinoma is a malignancy that may occur in different age groups and has the epidemiological features which are associated with oral carcinoma. Cowden's disease typically occurs in older people and causes fibroepithelial polyps. These polyps have less mobility and the location of oral involvement is different from the Heck disease (6). It's important to differentiate MEH from Condyloma Acuminatom because the clinical appearance of both diseases is similar and both caused by HPV; however, lesions in MEH are flatter and multiple. In addition, the lesions of MEH are mostly restricted to the buccal mucosa, lips, tongue, and other sites of oral mucosa (1, 6, 12), and since their self-limiting nature, most lesions do not require any treatments (1, 12, 13). However, if the lesions become chronic, enlarged, or recurrent, the disease requires substantial treatment (1). Nevertheless, the lesions of Condyloma Acuminatom are at the risk of transmission and malignancy (6). Various procedures have been recommended as the treatment plans, including topical treatments (for mild cases) and systemic treatments (in more severe cases). Due to the more common side effects of systemic therapy, topical treatment in children is preferable (1, 12, 13). Subsequently, our patient has family history and 2 out of five members of his family have MEH; therefore, the clinical features conducted us to the diagnosis of the Focal Epithelial Hyperplasia.

Various procedures of treatments were mentioned in the past studies such as topical cryotherapy as well as the use of vitamin A and sulfamides (1, 6). In the recent years, Laser therapy has been considered as a novel therapeutic approach. This method not only has better results but also reduces the risk of recurrence (8). Unfortunately, in spite of explanation and scheduling the surgical treatment for other lesions, our patient did not come after the diagnosis of the disease, because of being asymptomatic.

MEH is very rare in the Iranian population and only 18 cases of this disease have been reported mostly in the Iranian children (9-11) Since most of the reported cases were residents of Khorasan Province (Northeast, Iran) and also three reported cases were in an Afghan immigrant family group living in Iran, the racial factor and familial predominance should be considered for such patients in Iran (7, 9). However, we reported MEH in an adult man in Yazd (the central part of Iran).

Ultimately, Heck disease is very rare in the Iranian population and it is the 19th case of MEH in this country; therefore, timely diagnosis and prevention of its transmission have great importance. Consequently,

promotion of dentists' awareness of the diagnosis and appropriate referral to oral medicines specialists are recommended.

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Conflict of interest

The authors have no conflict of interest in this study.

Ethical statement

The patient gave his written informed consent to participate in the study and to have his photographs published for scientific purposes. The patient was informed about the nature, diagnosis, and treatment options of his condition and the possible risks and benefits of the procedure. The patient was also advised to follow up regularly and to report any changes or discomfort in his oral lesions.

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