Case Report

Oral Multifocal Epithelial Hyperplasia: An Unusual Entity

Karthikeya Patil, Mahima V Guledgud, CJ Sanjay, Bharathi Penumatsa[1]

Department of Oral Medicine and Radiology, JSS Dental College and Hospital, JSS Academy of Higher Education and Research, Mysore, Karnataka, [1]Department of Oral Medicine and Radiology, Care Dental College and Hospital, Guntur, Andhra Pradesh, India

Abstract

Multifocal epithelial hyperplasia (Heck's disease) is a rare, contagious, asymptomatic, and benign disease affecting the oral mucosa. It is usually seen in childhood and adolescence and is noted only in specific groups in certain geographical regions. The lesion is clinically characterized by the presence of multiple small papules or nodules in the oral cavity, especially on the labial mucosa, buccal mucosa, and tongue. Histopathology is necessary to confirm the diagnosis. Treatment remains unspecific, although surgical excision, CO_2 laser therapy, and topical antivirals are in use. We present a clinical case of a 65-year-old Indian male with multiple oral lesions that clinically and histologically resemble Heck's disease.

Keywords: *Heck's disease, multifocal epithelial hyperplasia, multiple oral lesions, papillomavirus*

Introduction

Multifocal epithelial hyperplasia (MEH) is a benign, rare, familial disorder, characterized by soft, circumscribed, multiple, sessile nodular elevations of the oral mucosa, primarily associated with human papillomavirus (HPV) types 13, 32.[1] Geographically, it is more prevalent in North, South, and Central American-Indians (Waimiri-Atroari Indians) and less common in caucasians.[2] Younger individuals with MEH exhibit multiple nodular lesions, whereas elderly patients exhibit few or even single lesions, which tend to be flat and papular.[3] Koilocytosis is constantly present in these lesions.[1]

Case Report

A 65-year-old immunocompetent male, with a personal history of smoking 20 beedis/day for the past 30 years presented to us with asymptomatic multiple papules on the buccal mucosa bilaterally and on the upper and lower labial mucosa ranging in size from 1 to 4 mm [Figure 1]. The lesions were grayish white, with no inflammatory component. The surfaces of these lesions were smooth, and they were firm in consistency. Lesions were nontender to palpation. His initial complaint was that of mobile teeth, being identified as chronic generalized periodontitis. Provisionally, the case for incidental finding was diagnosed with MEH and differential diagnosis included molluscum contagiosum, lymphangioma, condylama accuminatum, tuberous sclerosis, and Cowden's syndrome.

Routine hematological investigations revealed normal values. Tissue specimen was obtained from one of the papular lesions in the right posterior buccal mucosa for the histopathological study. Sections revealed stratified squamous epithelium with abrupt acanthosis, mild hyperkeratosis, and focal mild koilocytosis noted in the upper layers of the epithelium with perinuclear halo [Figures 2 and 3]. The histopathological picture was suggestive of MEH. Thus, a final diagnosis of Heck's disease was made.

Discussion

Dr. Archard Heck in 1965 described MEH (also known as Heck's disease or multifocal papilloma) as rare, benign lesions of the oral cavity. The frequency of Heck's disease is variable with a wide range of 0.002%–35% depending on the population and geographical region.[4] The disease has been reported so far from countries such as Argentina, Paraguay, Columbia, Ecuador, Venezuela, Brazil, Eisalvador, Guatemala, India, Israel, Egypt, Iraq, Greenland, Alaska, Britain, South Africa, and Tibet.[5]

The exact etiology of Heck's Disease is unknown, but tobacco, electrogalvanism,

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Address for correspondence:
Dr. Mahima V Guledgud, JSS Dental College and Hospital, JSS Academy of Higher Education and Research, Sri Shivarathreeshwara Nagar, Mysore - 570 015, Karnataka, India.
E-mail: mahimamds@gmail.com

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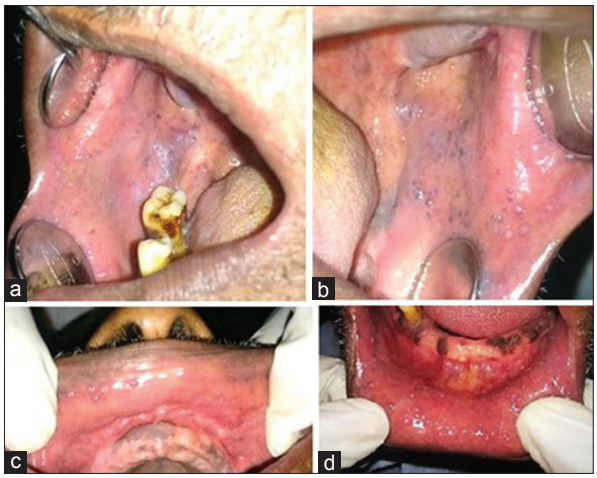

Figure 1: Clinical presentation: (a) Right buccal mucosa. (b) Left buccal mucosa. (c) Upper labial mucosa. (d) Lower labial mucosa

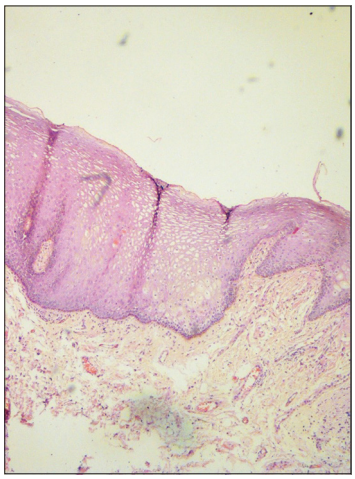

Figure 2: Photomicrograph showing abrupt severe acanthosis (H and E, ×100)

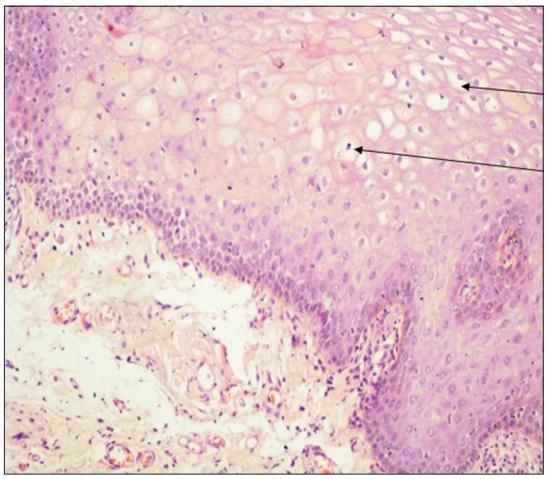

Figure 3: Photomicrograph demonstrating koilocytosis noted in the upper layers of the epithelium with perinuclear halo (arrows) (H and E, ×400)

and Vitamin A deficiency among others have been cited by few authors.[6] A viral etiology primarily HPV 13 and 32 has been suggested based on microscopic, electron microscopic, and immunofluorescent examination. Lower socioeconomic status, poor oral hygiene, crowded living conditions, malnutrition, and HIV infection seem to have additional risk factors.[7] Although familial distribution has been mentioned in previous studies, our patient did not report such findings in any of his family members. However, factors such as chronic smoking habit could have possibly attributed to the disease.

Heck's disease can manifest in either of the genders,[7] but studies report to have female predilection,[8] predominantly affects children and patients in the first and second decades of life with lower lip being the most commonly involved site.[7,8] Other sites include buccal, labial, and lingual mucosa. In our case, the disease presented at an unusual age of 65 years involving lesions in multifocal areas like buccal mucosa bilaterally and upper and lower labial mucosa.

MEH is clinically characterized by multiple circumscribed, discrete, sessile, soft, flattened or elevated, and round papulonodular lesions of the oral mucosa, with color similar to the adjoining oral mucosa. Rarely, lesions show a papillary surface change. Individual lesions are small measuring about 3–10 mm, and they frequently cluster together to give on a cobblestone appearance.[7] In areas with mechanical trauma, frictional keratosis will enhance the lesion and make it appear whiter. In our case, the region of the posterior right buccal mucosa showed grayish areas interspersed with white keratotic nonscrapable areas of frictional keratosis.

Differential diagnosis includes molluscum contagiosum, condyloma acuminatum, lymphangioma, Cowden syndrome, and tuberous sclerosis as they are known to have some clinical similarities such as lesion type, color, and location. Molluscum contagiosum present with multiple papules mainly seen in children and young adults, involving the neck, trunk, and genitalia. Condyloma acuminatum, clustered papules with the papillary surface, is seen in the floor of the mouth and ventral tongue because of urogenital contact with an infected partner. Oral lymphangiomas frequently involve anterior two-thirds of the tongue with a pebbly surface resembling a cluster of translucent vesicles. Cowden's syndrome encompasses with multiple hamartomas and tuberous sclerosis may be associated with cutaneous hypopigmentation, subungual fibroma, enamel defects, epilepsy, or mental impairment.

Clinical diagnosis of Heck's disease is confirmed by the histopathological examination. The prominent microscopic features consist of extensive acanthosis, parakeratin layering, degeneration of koilocytes, mitosoid cells, and elongated rete ridges some of which form horizontal anastomosing and other rete ridges form clubbing called "bronze age battle axe" or "clubs."[8] In our case, histologic H and E-stained sections imparted the presence of abrupt acanthosis and mild hyperkeratosis and with focal mild

koilocytosis. Although biopsy remains the gold standard in the diagnosis, molecular advances such as DNA *in situ* hybridization[8] for HPV types 1, 6, 11, 13, 16, 18, and 32 can be performed to arrive at a specific diagnosis of Heck's disease.

The management of MEH is not always mandatory since they are asymptomatic and tend to regress spontaneously. Only in those cases, treatment is essential, wherein areas of esthetics are concerned. The mainstay of therapy includes the removal of the lesion by excision biopsy, cryotherapy, CO_2 laser, electrocoagulation, or electrodessication. In cases of diffuse involvement, topical or systemic interferon is beneficial. Other treatment modalities include levamisole, topical podophyllin resin, or vitamins. In view of the multifocal presentation, CO_2 laser ablation was advised for the patient. However, the patient opted not to undergo treatment because of its asymptomatic nature. Further, long-term follow-up was advised to monitor the status of the lesions.

Conclusion

MEH, a rare case seen in Asian countries such as India, has an optimistic prognosis as it carries no malignant potential. Although commonly seen in younger age group, it can rarely be encountered in elderly patients as well. The disease, with its bizarre clinical course, poses a challenge to uninformed oral physicians. Hence, it is imperative to be aware of this condition so that early intervention is facilitated.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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