

Fully edentulous patient with multifocal epithelial hyperplasia: Report of very rare case

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World Journal of Biology Pharmacy and Health Sciences, 2022, 10(03), 027–030

Publication history: Received on 28 April 2022; revised on 04 June 2022; accepted on 06 June 2022

Article DOI: <https://doi.org/10.30574/wjbphs.2022.10.3.0082>

Abstract

Multifocal epithelial hyperplasia (MEH), also known as verrucae of the oral cavity, focal epithelial hyperplasia, Heck's disease, and multifocal papillomavirus epithelial hyperplasia, is a rare benign oral condition. It is most typically seen in the labial mucosa of younger people and rarely seen in middle-aged patients. Multiple small nodules or papules in the oral cavity, notably on the labial mucosa, buccal mucosa, and tongue, are clinically defined by this lesion. For precise diagnosis needs, clinical history, examination, and histopathologic findings. After months or years, spontaneous regression was commonly recorded; therefore, no treatment was required. Conservative surgical excision can be used for diagnostic and aesthetic reasons and recurrent trauma lesions. We present a very rare oral multifocal epithelial hyperplasia case involving the generalized attached gingiva of the upper and lower jaw in a fully edentulous patient in the Oral Specialized Clinic, Tehran, Iran.

Keywords: Attached Gingiva; Heck's Disease; Multifocal Epithelial Hyperplasia; Report

1. Introduction

Multifocal epithelial muscle hyperplasia (MEH) is a rare family member disorder characterized by soft, mucosal-colored, circumscribed, multiple, flattened papules or nodular elevations of the oral mucosa primarily associated with the oral mucosa with human papillomavirus (HPV) types 13, 32 [1,2]. HPV subtype 32 is more likely to cause disease in older people. HPV can impact both keratinized and non-keratinized intraoral surfaces [3]. Lower socioeconomic status, poor oral hygiene, crowded living conditions, malnutrition, and HIV infection are all risk factors [4]. Although it has been reported by other writers before, Dr. Heck published the first case report in 1965, and it is now known as "Heck's sickness" [5]. This condition usually appears in childhood or adolescence [6]. MEH is also more common in women than in men (75 percent versus 25 percent, respectively) [7]. The lower lip, buccal mucosa, and tongue are the most commonly affected areas [4,8]. It was first documented mostly among Native Americans, Eskimos, and South Africans, but reports have since spread to Nigeria, Polynesia, Puerto Rico, an adult female Caucasian, and minor ethnic groups throughout the world. Other ethnic groupings include Bolivia, Paraguay, Peru, Colombia, Mexico, and others. A precise diagnosis requires clinical history, examination, and histopathologic findings [9]. The present article reports a very rare case of oral multifocal epithelial hyperplasia involving the generalized attached gingiva of the upper and lower jaw in a fully edentulous middle-aged patient.

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2. Case report

In Tehran, Iran, a 59-year-old woman was hospitalized at an oral specialty facility. She complained of loose dentures and generalized multiple masses on the attached gingiva in the upper and lower jaw. Multiple pedunculated or nodular lesions with soft, smooth surfaces were seen in the intraoral examination. The tumors were the same color as the mucosa around them. These lesions were neither inflamed nor ulcerated, and they were completely asymptomatic. The diameter of the papules and nodules ranged from 1–2 mm. In the maxilla, the lesions were wider and sessile. (Figure 1) Lesions were not tender to palpation. According to the patient, the lesions have been present for two years and have not changed in size. Her family history was negative, and she had no systemic diseases. The patient had been wearing maxillary and mandibular complete dentures for five years.



Figure 1. Multifocal epithelial hyperplasia. (a) Multiple, pedunculated, soft, smooth surface papules and nodules involving mandible. (b) Multiple, wide, soft, sessile, smooth surface papules and nodules involving maxilla.

Routine hematological investigations revealed normal values. Due to clinical findings, oral squamous papilloma, warts, and irritation fibroma were suggested as differential diagnoses. In order to establish a final diagnosis, a biopsy was performed under local anesthesia on the mandible, and two papules were excised. An orthokeratinized stratified squamous epithelium lesion with acanthosis and extended rete ridges on histopathologic sections. A few koilocytes and mitosoid cells were detected in the epithelium's superficial layers. Sections of the salivary gland, blood vessels, hemorrhage, and muscle bundles were evident. There was no evidence of malignancy (Figure 2,3). Because of financial limitations, no polymerase chain reaction was done to recognize the type of virus. So, the final diagnosis of Heck's disease was established; considering the loose denture problem, surgery was determined as a suitable treatment choice. [Informed consent was obtained from the patient for the surgical procedure and necessary information for reporting this case].

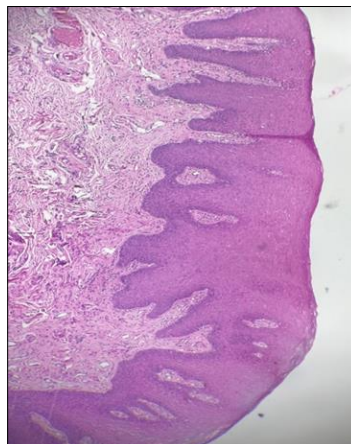


Figure 2. Orthokeratinized stratified squamous nuclei in epithelium with acanthosis, broad and elongated rete ridges. (×100 Magnification, H&E)

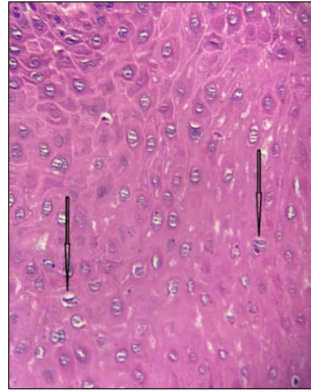


Figure 3. Mitosoid cells with altered superficial layer of epithelium (arrow). ($\times 400$ Magnification, H&E)

3. Discussion

MEH (also known as Heck's disease or multifocal papilloma) is a rare, benign oral cavity lesion first described by Dr. Archard Heck in 1965 [5]. The prevalence of Heck's illness varies greatly depending on the population's geographical region, ranging from 0.002 percent to 35 percent [10]. Individual lesions are tiny, usually 3–10 mm in diameter, and they frequently cluster together to create the appearance of cobblestones [4]. However, in our study, their dimensions were smaller than the previously specified usual size range because of the many lesions. MEH has been linked to HPV subtypes 13, 32, and 16, with reports of cross-reaction with subtypes 1, 6, 11, 16, 18, and 55 [4,11]. A recessive gene, vitamin K deficiency, tobacco chewing, the galvanic voltage from amalgam, unintentional biting, poor hygiene, poor socioeconomic position, and HIV infection have all been reported [4,12,13]. MFH frequently affects multiple family members [12] In our case, no indication of familial connection has been found. Bennett and Hinshaw showed that genetic variation, the human lymphocytic antigen-DR4 (HLA-DRB1*0404) allele, plays a vital role in this disease [4,14]. This condition usually occurs in the first two decades of life, and the underdeveloped immune system in younger patients has been explained [12] Heck disease can occur in both sexes, but studies report a female tendency [4]. The patient described in the present report had 59 years of age, an age above normal suffering, without any systematic disease. However, due to her advanced age, a compromised immune system might have predisposed her to MEH. The patient, in this case, was female, and this correlated with the majority of the studies. According to the study by Mosannen Mozaffari et al., the duration of lesions can vary from 2 months to 22 years [13]. The shorter duration correlates with this case. Differential diagnosis of MEH includes verruca vulgaris, papilloma, condyloma acuminatum, juvenile papillomatosis, irritation fibroma, verruciform xanthoma, Goltz-Gorlin, Cowden, MEN (multiple endocrine neoplasia), neurofibromatosis and tuberous sclerosis [13,15,16]. No treatment is necessary because spontaneous regression has been reported after months or years. Conservative surgical excision can be used for diagnostic and cosmetic reasons and for lesions that have been recurrently traumatized. In such circumstances, laser (CO₂), electrocautery, cryotherapy, scalpel surgery, sulfamide, topical interferon beta, systemic or intralesional interferon-alpha, or vitamin A were listed as treatment options [4,12,17,18]. Scalpel surgery was performed in this case, and the denture was referred to prosthodontics for relining.

4. Conclusion

A rare viral infection of the oral mucosa is known as multifocal epithelial hyperplasia (Heck's disease). It is usually detected in the labial mucosa of younger people, although it can also be found in the labial mucosa of middle-aged patients. Herein, we reported a generalized involvement of the attached gingiva in a fully edentulous, middle-aged patient by MEH.

Compliance with ethical standards

Disclosure of conflict of interest

The authors state that the publishing of this paper does not include any conflicts of interest.

Statement of informed consent

Informed consent was obtained from all individual participants included in the study.

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