Scholars Journal of Medical Case Reports
Abbreviated Key Title: Sch J Med Case Rep
ISSN 2347-9507 (Print) | ISSN 2347-6559 (Online)
Journal homepage: https://saspublishers.com

OPEN ACCESS

Dental Medicine

An Unusual Observation of Oral Multifocal Epithelial Hyperplasia in Elderly Man

Sarra Azzez[1*], Sarah Chkoundali[1], Ahlem Bellalah[2], Mehdi Khemiss[1], Mohamed Ben Khelifa[1]

[1]Dental Medicine Department, Fattouma Bourguiba Teaching Hospital, Monastir, Tunisia
[2]Department of Pathology, Fattouma Bourguiba Teaching Hospital, Monastir, Tunisia

DOI: 10.36347/sjmcr.2021.v09i12.028 | **Received:** 13.10.2021 | **Accepted:** 18.11.2021 | **Published:** 30.12.2021

***Corresponding author:** Sarra Azzez

Abstract **Case Report**

Multifocal epithelial hyperplasia or Heck's disease is uncommon, contagious, asymptomatic, and benign disease affecting the oral mucosa caused by some subtypes of human papilloma virus, especially subtypes 13 or 32. It affects mostly pediatric patients and adolescents in certain geographical regions and ethnic groups. Clinically, the lesion is characterized by the presence of multiple soft papules or nodules in the oral cavity. This paper reports a Tunisian case of 52-year-old man presented with Heck's disease. The diagnosis is based on both clinical aspect and histopathological features. Clinicians should be familiar with this rare oral disease, which can even affect elderly patients as reported in our case. Treatment is not mandatory and may be indicated when there is esthetic and/or functional impairment.

Keywords: Multifocal epithelial hyperplasia, Heck's disease, oral lesions, Human papillomavirus, Elderly patient.

INTRODUCTION

Published for the first time by Dr Heck and his team in 1965, multifocal epithelial hyperplasia (MEH), also known as Heck's disease, is a rare, asymptomatic, and benign disorder of oral mucosa [1, 2]. It's characterized by multiple, soft, sessile and well-circumscribed white-to-pinkish papules or nodular elevations on the oral cavity. It is localized most commonly on the labial and buccal mucosa, lower lip, tongue, and less often on the upper lip, gingiva, and palate [3]. Children and young adults are primarly affected without any difference between the sexes. This disease is apparently attributed to Human papillomavirus especially subtypes 13 and 32. However, cross-reaction with other subtypes has also been mentionned [1, 4]. Geographically, high frequency was found among indigenous population of North, South, and Central America and Eskimos from Greenland [4].

The aim of this paper is to report an unusual case of Heck's disease in elderly caucasian male.

CASE REPORT

A 52-year-old immunocompetent male was reffered to the Department of Dental Medicine of Fattouma Bourguiba Teaching Hospital of Monastir-Tunisia. His chief complain was recent appearance of painless multiple nodules on his buccal mucosa.

The patient denied any smoking habits or alcohol abuse nor the presence of similar lesion in his family members. His medical history and physical examination showed no abnormalities. Extraoral examination was non-contributory.

Intraoral examination revealed multiple, soft, sessile, non-inflammatory with smooth surface papules and nodules involving upper, and lower labial mucosa and left border of the tongue. These lesions were asymptomatic, white to pinkish in color and firm in consistency. Size of papules and nodules ranged from 3 to10 mm in diameter (Figure 1). Some of them were interfering with patient's speech and esthetic.

Figure 1: Intraoral examination showing multiple non-inflammatory papules and nodules involving upper, and lower labial mucosa and left border of the tongue

This case was suggestive of MEH and differential diagnosis included condyloma acuminatum, and Cowden's syndrome.

Routine hematological and serology investigations revealed normal values. An excisional biopsy of the larger nodule localized in the upper labial mucosa was carried out under local anesthesia.

Histopathological analysis showed stratified squamous epithelium with hyperkeratosis, parakeratosis, acanthosis and Hyperplasia of epithelial rete ridges. The superficial epithelial layers showed focal koilocytosis. The underlying connective tissue was without any atypia (Figure 2 and 3).

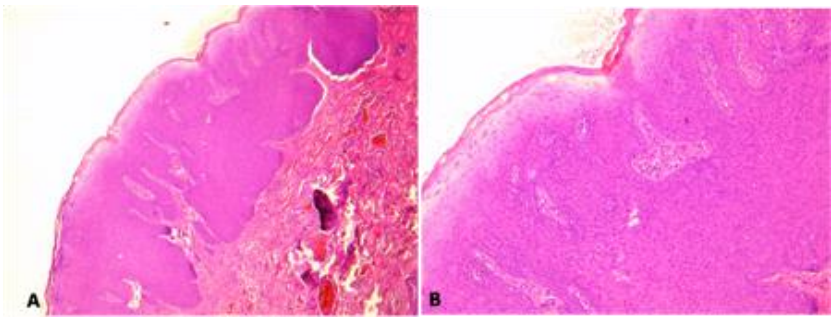

Figure 2: Acanthotic epithelium with parakeratosis (HE stain: A×40, Bx100)

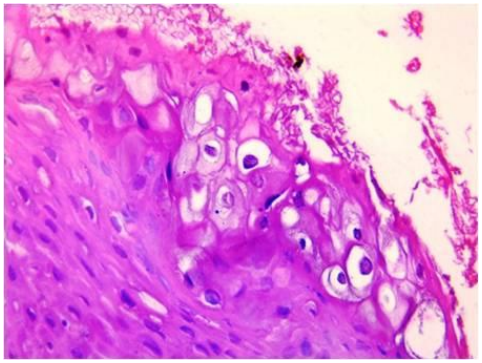

Figure 3: The superficial epithelial layers contain many koilocytes (HE stain x 400)

The patient presented after 10 days of removal of the labial nodule with spontaneous regression of the lesions (Figure 4). So, no treatment was required. Only regular clinical follow-up was recommended.

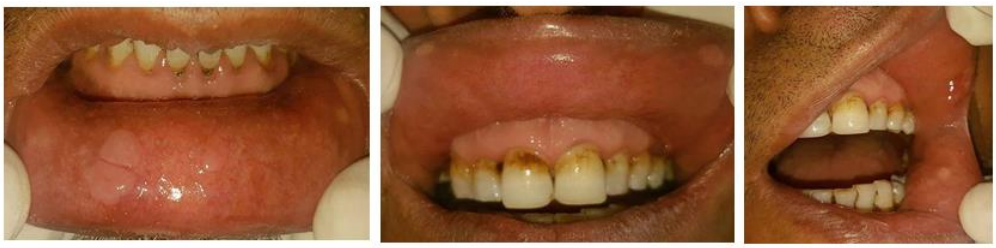

Figure 4: Spontaneous remission of the lesions after 10 days follow-up

DISCUSSION

MEH or Heck's disease is a rare benign disease, with typical clinical presentation and demographic profile. The frequency of Heck's disease is variable and accounts for 0.002%–35% depending mainly on the geographical region [5]. It's well-known in North, Central and South America and some parts of South Africa but it is an uncommon entity in North Africa [6].

There are several predisposing factors reported in the literature, such as genetic background, poor oral hygiene and low socioeconomic status, nutritional deficiency, and immunodeficiency [7]. Our patient had a poor oral hygiene but he does not belong to low socioeconomic groups. Although familial inheritance has been mentioned in previous studies, our patient did not report any similar lesions in his family members.

This uncommon, benign disease is induced by an infection with HPV type 13 or HPV type 32. Older age individuals tends to be affected by the subtype 32 of HPV, while the subtype 13 of HPV seems to affect similarly both young and old patients [8]. It predominantly affects children and young adults, in the first and second decades and in either of the genders, but some studies report a female predilection [8] with lower lip being the most commonly involved site.

Clinically, MEH classically represents as multiple, well-circumscribed, sessile, soft, flattened or elevated, papulonodular lesions of the oral mucosa, measuring between 3 and 10 mm, and they frequently manifest a cobblestone appearance by clustering together. Their colors are like the adjoining oral mucosa. In areas with mechanical trauma, frictional keratosis will add a whiter appearance to the lesions [2, 7]. In this paper, we described multiple mucosal lesions in a 56-year-old man located mainly at the lower and upper labial mucosa. It was reported that younger patients showed multiple nodular lesions, whereas elderly patients exhibited fewer flat and papular lesions [9].

The identification of Heck's disease is based both on the clinical aspect and the histopathological analysis. The mainstay of the microscopic features are acanthosis, parakeratin layering, degeneration of koilocytes, mitosoid cells, and elongated rete ridges. The histopathological examination of our case showed parakeratosis, acanthosis, rete pegs with a cytopathological feature very characteristic of HPV infection which is the presence of focal koilocytosis, a pathognomonic sign of MEH [10]. Those microscopic findings with clinical presentation were enough to establish the diagnosis. Although biopsy still be the gold standard, other molecular approach such as DNA in situ hybridization and Polymerase chain reaction genotyping for HPV types are of utmost interest to get a specific diagnosis of Heck's disease [10].

The differential diagnosis includes other HPV-induced lesions, such as condyloma acuminatum, verruca vulgaris and mucosal involvement of Crohn disease, Cowden syndrome and multiple endocrine neoplasia type 2B [6, 11].

The long-term evolution of MEH is unpredictable. It may remain stable for a long time, increase in size or spontaneously regress within months to years [12]. In our case, the patient had a spontaneous remission after few months.

The management of MEH is not always required given its asymptomatic nature and tendency to regress spontaneously. Only in those cases with functional and/or esthetic concerns, an intervention is recommended. Several treatment modalities have been proposed for MEH, like removal of the lesion by excision biopsy, cryotherapy, electrocoagulation, treatment with carbon dioxide laser, topical agents such as imiquimod, retinoic or trichloroacetic acid and immunostimulants especially systemic interferon [2, 6, 8].

Despite its association with HPV, the majority of the studies did not report any malignant potential of MEH. There is one study that has mentioned a malignant transformation in a case of MEH related to HPV 24 [8, 13].

CONCLUSION

MEH is uncommon, benign disease of the oral mucosa induced by HPV infection rarely seen in African countries such as Tunisia. Elderly patients could be affected eventhough it involved mainly children and young adults. Oral physicians should be vigilant and include this rare condition in the differential diagnosis when a patient is presented with multiple nodular lesions of the oral cavity. So, a rapid diagnosis could be established.

REFERENCES

1. Bascones-Martínez, A., Cok, S., Bascones-Ilundáin, C., Arias-Herrera, S., Gomez-Font, R., & Bascones-Ilundáin, J. (2012). Multifocal epithelial hyperplasia: A potentially precancerous disease?. *Oncology letters*, *3*(2), 255-258.
2. Patil, K., Guledgud, M. V., Sanjay, C. J., & Penumatsa, B. (2019). Oral multifocal epithelial hyperplasia: An unusual entity. *International Journal of Applied and Basic Medical Research*, *9*(4), 253-255.
3. Ledesma-Montes, C., Vega-Memije, E., Garcés-Ortíz, M., Cardiel-Nieves, M., & Juárez-Luna, C. (2005). Multifocal epithelial hyperplasia: Report of

nine cases. *Medicina Oral, Patología Oral y Cirugía Bucal (Ed. impresa)*, *10*(5), 394-401.
4. Said, A. K., Leao, J. C., Fedele, S., & Porter, S. R. (2013). Focal epithelial hyperplasia–an update. *Journal of oral pathology & medicine*, *42*(6), 435-442.
5. Puriene, A., Rimkevicius, A., & Gaigalas, M. (2011). Focal epithelial hyperplasia: Case report. *Stomatologija*, *13*(3), 102-104.
6. de Castro, L. A., de Castro, J. G. L., da Cruz, A. D. L., de Sousa Barbosa, B. H., de Spindula-Filho, J. V., & Costa, M. B. (2016). Focal epithelial hyperplasia (Heck's Disease) in a 57-year-old Brazilian patient: a case report and literature review. *Journal of clinical medicine research*, *8*(4), 346-350.
7. Neville, B. W., Damm, D. D., Allen, C. M., & Chi, A. (2015). Oral and Maxillofacial Pathology. 1st ed. South Asia: Elsevier India; p. 336.
8. Mansouri, Z., Bakhtiari, S., & Noormohamadi, R. (2015). Extensive focal epithelial hyperplasia: a case report. *Iranian journal of pathology*, *10*(4), 300-305.
9. Dos Santos, P. J. B., Bessa, C. F. N., De Aguiar, M. C. F., & Do Carmo, M. A. V. (2004). Cross-sectional study of oral mucosal conditions among a central Amazonian Indian community, Brazil. *Journal of oral pathology & medicine*, *33*(1), 7-12.
10. Bascones-Martínez, A., Cok, S., Bascones-Ilundáin, C., Arias-Herrera, S., Gomez-Font, R., & Bascones-Ilundáin, J. (2012). Multifocal epithelial hyperplasia: A potentially precancerous disease?. *Oncology letters*, *3*(2), 255-258.
11. Said, A. K., Leao, J. C., Fedele, S., & Porter, S. R. (2013). Focal epithelial hyperplasia–an update. *Journal of oral pathology & medicine*, *42*(6), 435-442.
12. Ledesma-Montes, C., Vega-Memije, E., Garcés-Ortíz, M., Cardiel-Nieves, M., & Juárez-Luna, C. (2005). Multifocal epithelial hyperplasia: Report of nine cases. *Medicina Oral, Patología Oral y Cirugía Bucal (Ed. impresa)*, *10*(5), 394-401.
13. Jablonska, S. (2000). Demonstration of HPV 24 in long-standing Heck's disease with malignant transformation. *European Journal of Dermatology*, *10*(3), 235-236.