

Multifocal Epithelial Hyperplasia: Clinical Diagnosis of an Uncommon Oral Mucosal Lesion. A Case Report

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ABSTRACT

Objective: Multifocal epithelial hyperplasia (MEH) or Heck's disease is an uncommon oral mucosa disease caused by the Human Papilloma Virus (HPV) especially subtypes 13 or 32. It is contagious with a tendency to be transmitted to other family members. There is geographic regional variation in the frequency of occurrence of this condition. A careful clinical examination is required to arrive at the diagnosis. Histopathological review however can help to confirm the diagnosis of MEH. Molecular biology techniques like PCR or ISH are useful in isolating the HPV subtypes involved. This report aims to highlight the clinical features of MEH for accurate clinical diagnosis by Dental and Medical practitioners

Case Report: A documentation of a case of a 6-year-old Efik, Nigerian male child with extensive multiple oral mucosa lesion of 2 years duration. The diagnosis of this case was based on the clinical presentation. The presence of multiple painless pink to whitish oral mucosa papules and nodules on the lips, labial and buccal mucosa was seen and a diagnosis of Multifocal Epithelial Hyperplasia was made.

Conclusion: We report a case of MEH, a rare benign oral mucosal lesion in a 6-year-old male of the Efik ethnic group in the south-south zone of Nigeria who presented with a 2year history of multiple lesions in the oral mucosa. It is important for Dental and Medical practitioners to be aware of this rare condition.

Keywords: Multifocal epithelial hyperplasia, diagnosis.

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INTRODUCTION

Focal epithelial hyperplasia also known as Heck's disease is now also presently referred to as the Multifocal Epithelial hyperplasia (MEH) due to its clinical oral presentation.^{1,2} World-wide, the frequency can vary from 0.002 to 35% depending on the population studied.² Although it is a relatively rare disease condition, in the Eskimo population, its prevalence varies from 7% to 36%.¹ As the name MEH indicates, the most prevalent clinical finding is the presence of multiple nodular lesions in the oral

mucosa.³ It is an uncommon benign disease seen to affect the oral mucosa.^{1,4,5} This disease was first reported in 1965 by Dr. Hecks and Archard.^{1,4} MEH is known to be caused by the human papillomavirus (HPV).^{1,4,5} It is a rare benign lesion of the oral mucosa produced by the subtypes 13 or 32 of HPV.^{4,6,7} A site-specific predilection for keratinized and non-keratinized surfaces has been observed in these two types of HPV, respectively. Moreover, the subtype 32 of HPV tends to cause the disease in the older age groups while the subtype 13 of HPV seems to be

equally involved in the development of the disease in both young and old patients.⁴ Over the past few years, immunohistochemical and In-situ hybridization studies (ISH) have shown MEH to be closely related to HPV-13 and HPV-32 mostly, while certain studies have also demonstrated the presence of HPV-1, HPV-6, HPV-11 and HPV-16 in their cases.^{8, 9} MEH is contagious, with a tendency for the virus to be transmitted from one affected family member to another.⁵ After Archard et al.¹⁰ described it in Native Americans in 1965, the majority of cases published since then has described this disease in various ethnic groups in Eskimos and North, South and Central American Indians.⁴ There is geographic variation in the frequency of occurrence of this disease from one geographical region to another.^{4,7} Multifocal epithelial hyperplasia has a variable female predilection.^{8, 11} It appears more commonly in children, but there are some communications in adults and it is characterized by the presence of multiple slow growing, papulo-nodular, elevated, and smooth surfaced, soft asymptomatic lesions. Usually, they are sessile, round or oval, well defined nodules with colour similar to the normal mucosa, measuring from 0.1 to 0.5 cm in diameter. Coalescence of several small swellings gives rise to multinodular lesions of several centimeters.^{3, 12, 13, 14} They are predominantly found on the lower lip, buccal mucosa and tongue, and less often on the upper lip, gingiva and palate.^{5, 15} The presence and persistence of this disease can result in psychosocial problems.^{16, 17} The diagnosis is based on clinical grounds, and treatment is usually unnecessary since most of lesions regress spontaneously. The management of MEH lesions is only required for aesthetic or functional purposes.^{11, 18} To the best of our knowledge, no case of MEH has been reported in our own environment. Therefore this paper is aimed at reporting a case of a MEH, a rare entity seen in our environment and to also highlight the clinical features of this condition for accurate clinical diagnosis by Dental and Medical practitioners who will in turn allay the fears of the patients and counsel them on the benignity of this lesion.

CASE REPORT

A 6-year-old Nigerian boy from the Efik ethnic group in the south-south zone of Nigeria presented to the Vincintore, Dental clinic, Calabar with multiple oral mucosal lesions in the mouth of 2 years duration. The lesions were painless but there were rare occasions of traumatic injuries to the lesions arising from biting when the patient masticates. The patient appeared

healthy looking and his past medical and dental history were non-contributory. No abnormalities were observed on extraoral examination. Intraorally, there were widespread numerous multiple soft, sessile, smooth to ragged papular and nodular, pink to whitish projections on the oral mucosa some of which were coalescing to form larger nodules. These multiple lesions involved the lower and upper lip, the buccal mucosa and around the tip of the tongue. The size of the lesions ranged from 2mm to 1cm (Figure 1 and 2).

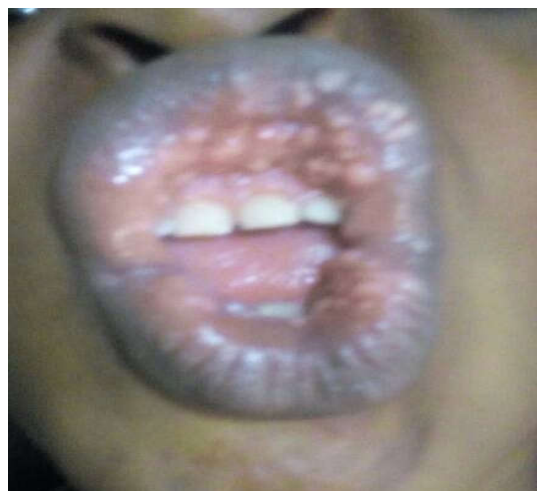


Figure 1. Extensive multiple oral mucosa papules and nodules on the lips

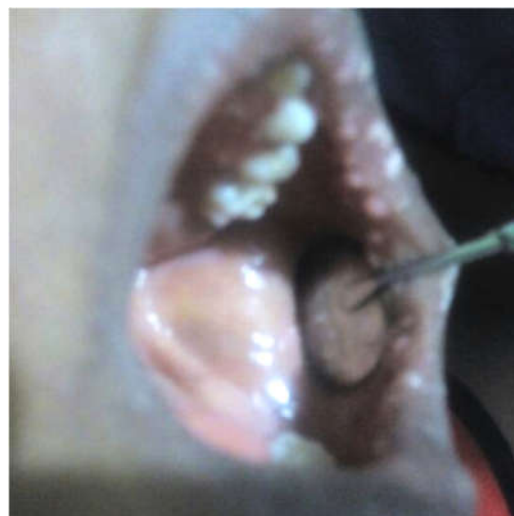


Figure 2: Extensive multiple oral mucosa papules and nodules on the lips and left buccal mucosa

The patient is an only child of the parents. The parents appeared visibly anxious as a result of the

presence of this lesion in their child. Counseling was done to reassure on the benign nature of the condition and the tendency to regress spontaneously with time. No surgical intervention was done because the history of biting on the lesions is infrequent. Patient was however placed on retinoic acid 400 microgram daily. Review of the patient after 3 months showed no remarkable change in the clinical presentation of the lesions. This was however expected since resolution of MEH usually takes months to years. Patient is still undergoing follow-up review.

DISCUSSION

Several authors have reported on MEH, a rare oral lesion caused by the virus HPV, mostly the subtypes 13 and 32.^{4, 6, 7, 13} The diagnosis of this disease is not usually challenging. This is due to its clinical presentation and so quite easily diagnosed. Therefore, a good demographic history including communal lifestyle and a careful and comprehensive clinical examination will help in arriving at a diagnosis of MEH.^{5, 19} Hence information regarding communal way of life, including characteristic sharing of food, personal objects and lack of hygiene, are helpful.^{5, 19} The clinical presentation in our patient is similar to that observed by previous authors^{3, 12-14} who reported multiple, painless, non-tender papules and nodules located in the lips, buccal mucosa and tongue occurring more frequently in children. Hence making a diagnosis of MEH in this 6-year-old child was not challenging for us because of the manifestation of these characteristic clinical features. Also, clusters of the lesions seen in this case forming larger lesions is similar to the reports by Ledesma-Montes et al.¹² and Hashemipour et al.²⁰ that these lesions may frequently be clustered giving a cobblestone appearance. The persistence of this lesion in this patient for 2 years agrees with reports that MEH may last for several months, or years, before running out its course.^{4, 10} De Castro et al.¹¹ and Bascones-Martínez et al.⁹ stated that MEH can be diagnosed mainly on the basis of clinical observations. The diagnosis of MEH in our case report was based on clinical findings.

However, MEH can be further diagnosed by histopathological evaluation and the finding will reveal proliferation of oral squamous epithelial cells with characteristics of viral infection composed of the presence of koilocytes having perinuclear halo helps to strengthen the diagnosis.^{3, 11, 20} In our environment, which is a resource limited economy,

isolating the viral strains in a patient with MEH is a challenge. This will require advance investigation technique like PCR which is a useful tool to identify the viral etiology of MEH lesions because it is a rapid and sensitive method.^{5, 11, 21} Borborema-Santos et al.⁵ in their study of 5 cases of MEH using PCR observed that a wide range of viral diversity can be identified with this method. They reported that the presence of HPV once detected, sequencing of PCR products is important to establish which viral type was actually the etiologic agent of MEH and in their series HPV-13 was implicated in all their 5 cases. Also, In-situ DNA hybridization has also proven to be highly useful in identifying the types of HPV involved.^{9, 14}

Our patient is presently under follow-up since MEH disease has a tendency to undergo regression with time, although it can persist for many years. In this case counseling was done to allay fears and to re-emphasize the benignity of this disease. Treatment is not always necessary and since trauma to the lesion was not frequent in this case, surgical intervention was the least of our consideration. Surgical treatment is only advised when lip involvement is causing aesthetic problems or lesions that are constantly traumatized by biting. Treatment may also include scalpel surgery, cryotherapy, CO₂ laser surgery, electrocoagulation, and conservative treatment using retinoic acid or interferon, an immunostimulant.^{5, 11, 15, 17, 18, 20}

CONCLUSION

In this case, we reported a case of MEH, a rare benign oral mucosal disease in a 6-year-old male of the Efik ethnic group in the south-south zone of Nigeria who had multiple lesions in the oral mucosa of 2 years duration. Dental and Medical practitioners must be aware of this rare condition so as to facilitate accurate diagnosis, adequate treatment if necessary, counseling and follow up.

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