RARE CASE OF MULTIFOCAL EPITHELIAL HYPERPLASIA OF ORAL MUCOSA

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ABSTRACT:

Introduction: Multifocal papilloma virus epithelial hyperplasia (MPVEH) is a rare disease of the oral mucosa associated with the human papilloma viruses (HPV) 13 and 32. It occurs at any age, but children have the highest incidence.

Purpose: To present a case of oral multifocal papilloma virus epithelial hyperplasia with palatal tumour and multiple metachronic primary cancers.

Material and methods: The medical history shows a 60 years female patient with complaints of asymptomatic mass on the palatal mucosa, with a two years’ development. The patient has been treated surgically for previous thyroid cancer, endometrial cancer and cancer of the mammary gland. An incisional biopsy has been performed in her left buccal mucosa 10 years ago and histopathological examination confirmed the diagnosis of MPVEH.

The palatal tumour was removed surgically by electroexcision and the histological result was pleomorphic adenoma of a small palatal salivary gland.

Conclusion: Papillomatosis is considered as a benign neoplasm with an unpredictable course and with a low probability of malignancy. We report of an unusual case of MPVEH persistent for more than 30 years without regression and with significant impact on a patient’s quality of life.

Key words: Multifocal papilloma virus epithelial hyperplasia, palatal tumour, multiple primary tumours.

INTRODUCTION:

Multifocal epithelial hyperplasia (MEH) is also described as verrucae of the oral cavity, focal epithelial hyperplasia, Heck’s disease, and multifocal papilloma virus epithelial hyperplasia. The condition was first noted in the Spanish literature by March in 1881 and later by Stern [7] and Helms.[9] In 1961, Heck came across the condition again and the earliest English publications were by Archand et al.[1] and Witkop and Niswander [15] in 1965. Heck noted multiple papules spread throughout the oral mucosa in an 11-year-old Navajo girl.[1, 4] A series of 19 pediatric patients with intraoral papules was reported, subsequently with the authors coining the term “focal epithelial hyperplasia” to describe the intraoral papules found in affected Indian children from the United States and Brazil and an Eskimo child from Alaska.[1] In 1966, Hettwer and Rodgers[8] noted focal epithelial hyperplasia in a Polynesian girl and named the condition “Heck’s disease” with the eponym enduring in the dental literature; however the name of “multifocal epithelial hyperplasia” is the most appropriate, as it describes the major features of the disease (the presence of multiple lesions widespread on the oral mucosa and the microscopically prominent hyperplasia of the oral epithelium Multifocal epithelial hyperplasia (MEH) is a rare disease of the oral mucosa usually associated with the human papilloma viruses (HPV) 13 and 32.[11] It presents as multiple nodular or papular lesions with a sessile base, ranging in diameter from 0.1 cm to 1.0 cm, frequently coalescing. Lesion colour varies from red to white, depending on the presence and degree of frictional keratinisation. The condition occurs at any age, but children have the highest incidence. Multiple epithelial hyperplasia appears mainly in childhood and adolescence between 3 and 18 years of age and tends to spontaneously regress, although it can persist for many years. The condition is rare in people living in urban society, with most cases noted in tribal communities from remote Greenland and North, Central, and South America.[9] It is more common among females.[14] The lesions are usually painless, unless traumatized, and are frequently found on the buccal mucosa, lower lip, and commissures but also found on the mucosa of the upper lip, tongue, hard palate, and gingival. The etiological factors and pathogenesis of MEH are speculative, but mainly associated with the human papilloma viruses (HPV) 13 and 32 type.

The histologic appearance of epithelial hyperplasia varies with basal epithelial club-shaped projections that occasionally anastomose horizontally (“bronze age battle-axe” appearance), koliocytes, and other intracellular changes indicative of viral infection such as hyperchromatism and enlargement of the nuclei. Human papillomavirus 13 and 32 DNA have been consistently detected in these lesions. The
degenerating epithelial cells resemble cells in various phases of mitosis and are called mitosoid cells.[7] These are mostly found in the upper part of the epithelium and in younger lesions but are not consistently present, with dilated capillaries and some lymphocytes evident in the connective tissue.[12]

The differential diagnoses include condyloma acumminatum, florid oral papillomatosis, Cowden’s syndrome, Crohn’s disease, Darier disease, focal dermal hypoplasia (Goltz syndrome), and white spongy naevus.[7]

CASE REPORT

A 60 years old female reffered to our department of Oral and Maxillofacial surgery with a mass on palatal mucosa with a two years development and with history of multiple oral masses of papules lesions since 30 years. (Fig. 1) The examination revealing several asymptomatic, exophytic, pink, sessile, smooth-surfaced nodules on the dorsal surface of the tongue, lingual surface of the lower lip, gingiva of upper and lower jaw and buccal mucosa. (Fig. 2, Fig. 3).

Multiple papules and nodules merging into “intestine”- like mucosa could be seen on the tongue, retrocommissural area, inside upper and lower lip and, gingival, buccal mucosa, bilaterally. The lesions were normal mucosa coloured, raised, smooth and with crypts inside.

Medical history of the patient includes multiple primary cancers.

The patient has been treated surgically for previous - thyroid cancer, - endometrial cancer and -cancer of the mammary gland.

An incisional biopsy has been performed in her left buccal mucosa 10 years ago and histopathological examination confirmed the diagnosis of Multifocal Human Papilloma Virus Epithelial Hyperplasia- MPVEN.

In our department of Maxillofacial surgery, University Hospital ‘St. Anna’, Sofia the palatal tumour was removed surgically by electroexcision and the histological result was cystic adenoma of a small palatal salivary gland (#3043/31.03.2013). The surgical border was 0.5 cm and clinically and histologically the borders were clean.(Fig. 4) The postoperative defect was covered by gauze, soaked with Iodasept unguentum. (Fig. 5) Intraoperative and postoperative period was without any complication. The gauze was removed 7 days after surgery, and the postoperative wound was healing secondary. The patient was follow up.

DISCUSSION

Although MEH is predominantly found in certain populations in America and Greenland,[7] it can be seen in other geographic regions like sporadic cases. Host factors such as immune suppression (HIV infection- unusual HPV types), genetic predisposition, malnutrition, and hygiene are key putative etiological factors.[4, 6] It has been proposed that a combination of these factors increases an individual’s risk for MEH.[9] A genetic predisposition has been proposed based on familial occurrences and ethnic predilections.[13] There have been multiple reports of the disease occurring in different ethnic groups living in poverty such as Indian communities from North, Central, and South America as well as in Inuit from Greenland.[15] A genetic predisposition may render an individual’s immune system susceptible to acquiring certain types of HPV infection.[5] The clinical course of MEH is benign and associated with HPV serotypes with minimal carcinogenic risk; therefore, aggressive therapy is usually not indicated.[14, 10] Removal of MEH lesions is indicated in patients who have significant functional disturbance or aesthetic concern. Current therapeutic options include surgical excision, cryosurgery, electrocoagulatory (carbon dioxide) lasers, interferon-__ injections, topical application of interferon-__ or podophyllin, topical and systemic retinoids, vitamins,[3] and diode laser removal.[2] Ideal treatment for symptomatic lesions in pediatric patients is one that is noninvasive and has minimal or no systemic side effects. The patient in this case has been informed of the benign nature of the lesions and close monitoring of the patients will continue.

CONCLUSION:

Papillomatosis is considered as a benign neoplasm with an unpredictable course and with a low probability of malignancy. We report of an unusual case of MEH persistent for more than 30 years without regression and with significant impact on a patient’s quality of life.

REFERENCES:

Fig. 2. Papilomatosis of oral mucosa- tongue, buccal mucosa and gingiva of lower jaw

Fig. 3. Palatal tumour

Fig. 4. Postoperative defect after electroexcision of tumour

Fig. 5. Postoperative defect covered with sutured gauze, impregnated with Iodasept unguent.


10. March CJ. Multiple papillary tumours of the labial, buccal and glossal mucous membrane. Dental Cosmos 1881;23:165.


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