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CASE REPORT

Focal epithelial hyperplasia in a human immuno-deficiency virus patient treated with laser surgery

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Abstract

Focal epithelial hyperplasia (FEH), or Heck's disease, is a rare disease of the oral mucosa; it is mostly found in children or young adults who are immunosuppressed and who live in regions with low socioeconomic status. It is characterized by asymptomatic papules on the oral mucosa, gingiva, tongue, and lips. Healing can be spontaneous, and treatment is indicated if there are aesthetic or functional complications. Human papillomavirus, especially genotypes 13 and 32, has been associated with FEH and is detected in the majority of lesions. Histopathologically, FEH is characterized by parakeratosis, epithelial hyperplasia, focal acanthosis, and fusion and horizontal outgrowth of epithelial ridges. A 37-year-old male patient was referred to the Department of Oral and Maxillofacial Sciences at the Sapienza University of Rome, complaining of numerous exophytic lesions in his mouth. He stated that the lesions were not painful but he had experienced occasional bleeding after incidental masticatory trauma. He had received no previous treatment for the oral lesions. His medical history revealed that he was human immuno-deficiency virus positive and was a smoker with numerous, asymptomatic oral papules clinically and histologically corresponding to FEH. The labial and buccal mucosa were especially affected by lesions. Surgical treatment was performed using a 532-nm potassium titanyl phosphate laser (SmartLite, Deka, Florence, Italy) in continuous mode with a 300 μm fiber and power of 1.4 W (power density 1980.22 W/cm^2). After anesthesia without vasoconstrictors, the lesions were tractioned with sutures or an Allis clamp and then completely excised. The lesions were preserved in 10% formalin for histological examination, which confirmed the clinical diagnosis of FEH. In this case, the laser allowed excellent control of bleeding, without postoperative sutures, and optimal wound healing.

Key words: Lasers; Focal epithelial hyperplasia; Mouth; Human immunodeficiency virus; Oral pathology

Core tip: Focal epithelial hyperplasia (FEH), or Heck's disease, is a rare disease of the oral mucosa, characterized by asymptomatic papules in the oral cavity. Human papillomaviruses have been associated with FEH and have been detected in the majority of lesions. Histopathologically, FEH is characterized by parakeratosis, epithelial hyperplasia, and acanthosis. Here, the case of a 37-year-old male patient, human immuno-deficiency virus-positive, smoker, with numerous asymptomatic oral papules clinically and histologically corresponding to FEH is described. Surgical treatment was performed using a 532-nm potassium-titanyl-phosphate laser. In this case, the laser allowed excellent control of bleeding without postoperative sutures and optimal wound healing.

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INTRODUCTION

Focal epithelial hyperplasia (FEH), or Heck's disease, is an uncommon, benign disease of the oral mucosa; it is mostly found in children and young adults. It has also been described in some Native American communities in North and South America, as well as in Eskimos of Greenland[1,2]. FEH is characterized by numerous nodules or papules, usually painless, with sizes that vary from 1 mm to 1 cm, that are mainly found on the lips, buccal mucosa, tongue and palate[3]. Human papillomavirus (HPV) has been detected in the lesions with both electron microscopy and DNA testing[4,5]. The most frequently involved viruses are HPV 13 and HPV 32[5]. The case of a young HIV-positive adult affected by FEH is reported below.

CASE REPORT

An African male patient, 37-year-old, was referred to the Department of Oral and Maxillofacial Sciences at the Sapienza University of Rome, complaining about numerous exophytic lesions in his mouth. He stated that the lesions were not painful but that he had experienced occasional bleeding after incidental masticatory trauma. He had not received previous treatment for the oral lesions. His medical history revealed that he was human immunodeficiency virus (HIV)-positive. The diagnosis of HIV was made 2 years previously. He was, at the time of the visit, in treatment with lopinavir and ritonavir (Kaletra, Abbott Italy, Campoverde di Aprilia, Italy), emtricitabine and tenofovir (Truvada, Gilead, Foster City, CA, United States), and in prophylaxis with sulfamethoxazole and trimetoprim (Bactrim, Roche, Milan, Italy). He also smoked 10 cigarettes per day, but he did not make use of alcoholic drinks.

According to the classification of the Centers for Disease Control, he was classified as stage A3.

Extraoral examination did not reveal any signs of other diseases. Intraoral examination showed the presence of 17 sessile, soft, normochromic lesions in the oral cavity. The lesions were localized on the lower lip and the buccal mucosa, on both sides (Figure 1). After the examination, a diagnostic hypothesis of FEH was assumed. Excisional biopsy was performed on one of the lesions to confirm the diagnostic hypothesis. The biopsy was performed using a potassium-titanyl-phosphate (KTP) laser with a wavelength of 532 nm (SmartLite, Deka, Florence, Italy) in continuous mode with a 300 μm fiber, power 1.4 W (power density 1980.22 W/cm^2). After anesthesia without vasoconstrictors (Mepivacaina Pierrel 30 mg/mL, Pierrel, Milan, Italy) the lesions were tractioned with sutures or an Allis clamp and then completely excised (Figure 2).

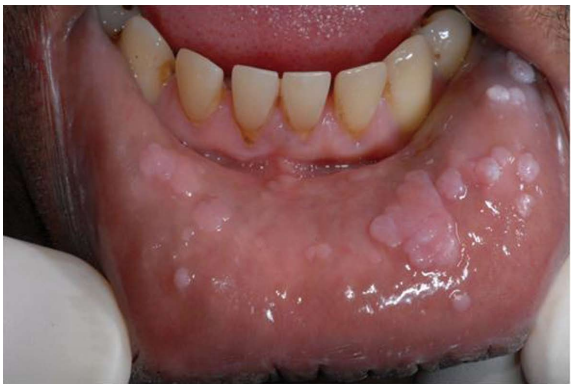

Figure 1 Clinical aspect of the focal epithelial hyperplasia lesions on the lower lip mucosa.

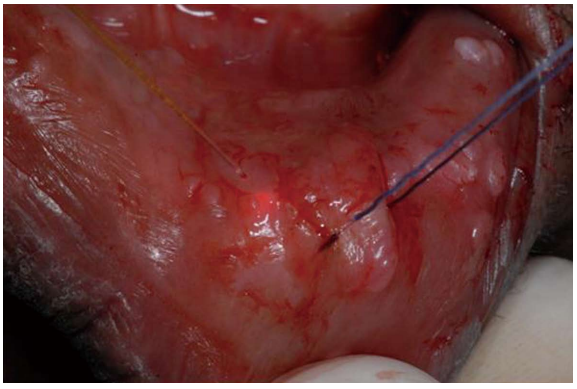

Figure 2 Laser excisional biopsy of one of the lesions on the lower lip.

The specimens were preserved in 10% buffered formalin for histological examination, which confirmed the clinical diagnosis of FEH.

In this case, the laser allowed excellent control of bleeding, without postoperative sutures, and optimal wound healing. After the first excisional biopsy, all of the lesions were surgically removed, in several steps, using the same operative approach. The patient was monitored with follow-up visits for one year, during which no recurrence of the pathology was observed (Figure 3), and he was asked to return if any lesions reappeared in the future.

DISCUSSION

FEH is a rare condition in Italy and Europe. Here, we describe the case of an immunosuppressed African man with FEH. This pathology has been extensively described among native South and North American populations[1,6-8]. It seems that there could be a genetic predilection for FEH, as cases seem to be limited to specific ethnic groups in certain geographic regions[1,6]. Ledesma-Montes *et al*[7] suggested a series of factors that could contribute to the development of FEH: poverty, genetic predisposition (ethnic factors), and a deficient hygienic lifestyle. FEH lesions are most frequently localized on the buccal mucosa, lip, tongue, and commissures; the retromolar area, palate, and mouth floor are rarer local-

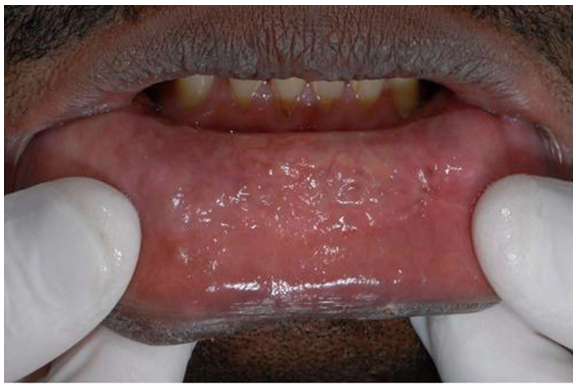

Figure 3 Clinical aspect of the healing after excision of the lesions on the lower lip, 1 yr after treatment.

izations[7]. FEH must be included in differential diagnosis with several pathological conditions that can be observed in the oral cavity[8], namely, condylomata acuminata, verrucous carcinoma, inflammatory fibrous hyperplasia, inflammatory papillary hyperplasia, and verruciform xanthoma[3]. Condyloma may appear similarly because it is caused by the same virus, but FEH lesions are more numerous and flatter, with typical localizations (buccal mucosa, lip and tongue)[9]. Verrucous carcinoma is a malign neoplasm that usually occurs in a different age group, usually in the sixth decade of life, with epidemiological characteristics that are similar to other oral carcinomas[10].

The last three pathologies are reactive lesions that usually occur with an irritating stimulus[9]. The diagnosis of FEH can be performed on the basis of clinical observation and can be confirmed by histological examination[11-13].

The histological features of this disease are parakeratosis, acanthosis, elongation of rete ridges, some of which may be anastomosed (the so-called "bronze age battle-axe" appearance[14], and usually koilocytes, as well as other cellular modifications that can indicate viral infection[5,14]. Cells with nuclear degeneration, called mitosoid cells, can also appear[15]. FEH can be associated with HIV infection, although the relationship between these conditions has not yet been completely clarified[12]. Suppression of the immune system leaves the patient vulnerable to opportunistic infections, including HPV infections[12].

There is no agreement in the literature on the potential malignant transformation of FEH lesions in immunocompromised patients. Moerman *et al*[12] stated that FEH lesions may have a high risk of malignant transformation in immunocompromised patients. Durso *et al*[13] tended to consider FEH a benign condition and stated that to date, no research has demonstrated the potential for malignant transformation of FEH lesions with HPV 13 and 32 subtypes. Only one case of malignant transformation of FEH caused by HPV type 24 has been reported[16]. Further studies are required to clarify this point.

Several therapeutic approaches have been proposed throughout the years. Some authors advise against removing the lesions because spontaneous regression can be observed[17], especially in children. Steinhoff *et al*[18] successfully treated FEH with topical applications of interferon beta. Other described methods include scalpel surgery, electrocoagulation, electrodessication, cryosurgery, and laser surgery[19,20]. In this case, laser surgery allowed excellent control of bleeding, without postoperative sutures, and optimal wound healing. Moreover, histological analysis is always possible with laser surgery when the proper parameters and correct surgical technique are used[21].

COMMENTS

Case characteristics

A 37-year-old male with a history of human immuno-deficiency virus (HIV) infection presented with numerous asymptomatic lesions in the oral cavity.

Clinical diagnosis

Seventeen sessile, soft, normochromic lesions on the lower lip and the buccal mucosa, on both sides.

Differential diagnosis

Condylomata acuminata, verrucous carcinoma, inflammatory fibrous hyperplasia, inflammatory papillary hyperplasia, verruciform xanthoma.

Laboratory diagnosis

Cluster of differentiation 4 receptors (CD4) 129/μL; HIV RNA < 37 copies/mL; metabolic panel and coagulation within normal limits.

Pathological diagnosis

Histological examination revealed parakeratosis, acanthosis, presence of koilocytes and mitosoid cells.

Treatment

Excisional biopsy with a 532-nm potassium titanyl phosphate (KTP) laser.

Related reports

Focal epithelial hyperplasia (FEH) can be associated with HIV infection, although the relationship between these two conditions has not yet been completely clarified; most likely, suppression of the immune system leaves the patient vulnerable to opportunistic infections, including HPV infections.

Term explanation

KTP lasers are powerful tools for oral surgery and oral pathology, as are other types of lasers.

Experiences and lessons

Oral lesions can be a manifestation of more complex systemic diseases; advanced surgical techniques can be useful tools in the management of multiple oral viral lesions.

Peer review

This paper is worthy of publication as an interesting case report of uncommon oral mucosa disease in HIV infected patient. Particularly, an information of efficiency of KTP laser surgery in treatment of Heck's disease will be useful for clinicians.

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