CASE REPORT

Focal Epithelial Hyperplasia Treated With Cryosurgery: A Case Report

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ABSTRACT

Focal Epithelial Hyperplasia Treated With Cryosurgery: A Case Report

Focal epithelial hyperplasia (FEH) is a rare, benign disease of the oral mucosa, which is caused by human papillomavirus (HPV) genotypes 13 and 32 with probable genetic susceptibility.

In this case report, we present the clinical, histopathological features and treatment of lesions observed in the oral mucosa of a 16-year-old female patient. Although FEH does not require any treatment in most cases, various treatments are available for patients with aesthetic concerns or complaints of functional impairment due to constant biting on the lesions. Cryosurgery was preferred for this patient with multiple lesions varying in size from 0.2 to 1.5 cm because of its many advantages, and considerable improvement was observed in the lesions.

KEYWORDS

Cryosurgery, Focal Epithelial Hyperplasia, Heck's Disease, Human Papillomavirus

ÖZ

Kriyocerrahi ile Tedavi Edilen Fokal Epitelyal Hiperplazi: Bir Olgu Sunumu

Fokal epitelyal hiperplazi (FEH) human papilloma virus (HPV) 13 veya 32 genotipleri tarafından üretilen, genetik geçiş izlenebilen oral mukozanın nadir, benign bir hastalığıdır.

Bu vaka sunumunda 16 yaşındaki kadın hastanın oral mukozasında gözlenen lezyonların klinik, histopatolojik özellikleri ve tedavisi sunuldu. FEH'de çoğu kez tedavi gereksinimi olmasa da estetik ve sürekli ısırma şikayetleri sonucu çeşitli tedaviler tercih edilebilir. 0.2 ile 1.5 cm arasında değişen çoklu lezyonlar izlenen bu vakada pekçok avantajları nedeniyle kriyocerrahi tedavisi tercih edildi ve lezyonlarda belirgin bir iyileşme gözlendi.

ANAHTAR KELİMELER

Fokal Epitelyal Hiperplazi, Heck Hastalığı, Kriyocerrahi, İnsan Papilloma Virüs

INTRODUCTION

Focal epithelial hyperplasia (FEH), also known as multifocal papilloma or Heck's disease, is a rare, benign lesion of the oral mucosa, associated with human papillomavirus (HPV) infection, mainly with genotypes 13 and 32 ([1]). The first case report was published by Dr. Heck and his colleagues in 1965 ([2]). Globally, FEH is rare; however, it is highly prevalent among Inuits and Indians residing in Central , North and South America, descendants of Khoi-San in South Africa and Eskimos from Greenland and North Canada ([3]).

The disease is more common in younger age groups and there is sometimes familial predisposition. This may be related to HPV transmission among family members. Risk factors for FEH include crowded living conditions poverty, malnutrition and poor oral hygiene ([4]). Clinically, FEH is characterized by the presence of numerous, soft, sessile papules and nodules with a diameter ranging from 1 to 10 mm, similar in color to the surrounding mucosa and usually the lips, buccal mucosa and the tongue are affected ([3,5]).

On histopathological examination, epithelial hyperplasia, epidermal acanthosis and parakeratosis, elongation and thickening of rete ridges, as well as large, vacuolated, glycogen-rich multinuclear cells and characteristic figures and koilocytic squamous epithelial cells are observed ([6,7]).

The diagnosis is based on clinical manifestations, and treatment is usually unnecessary since most lesions resolve spontaneously and do not exhibit tendency to malignant transformation. Treatment may only be required for aesthetic or functional purposes ([3,5,8]). Treatment options include topical imiquimod, intralesional and systemic interferon, CO_2 laser, cryosurgery, and electrosurgery ([7]). Among these modalities, cryosurgery is a treatment method that causes tissue necrosis by freezing the cells in the tissue to which it is applied ([9]).

With this case report, we aimed to present information about the clinical and histopathological findings of FEH and the beneficial therapeutic effects of cryosurgery on the lesions when used as a treatment modality for FEH.

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CASE REPORT

A 16-year-old healthy female patient with no systemic disease presented to the Department of Oral and Maxillofacial Radiology Cumhuriyet University, Faculty of Dentistry due to aesthetic concerns. On intraoral examination, multiple, swollen, sessile lesions with normal mucosa color were detected by palpation on the right/left cheek mucosa and lower/upper lip mucosal surfaces (Figure 1).

Figure 1.

Images of the right/left buccal mucosa (top) and lower/upper lip mucosa (bottom) of the patient

These lesions, ranging from 0.2 to 1.5 cm in diameter, were asymptomatic, with no signs of inflammation or ulceration. The lesions did not interfere with chewing. Intraoral lesions were present for three years and caused aesthetic concerns in the patient. Radiographic examination was unremarkable and no pathological lymph nodes were found on extraoral examination.

The patient reported that similar lesions were present her mother and elder sister and disappeared spontaneously over time. The patient and her family were informed about the procedures to be performed and their approval was obtained. Some of the lesions were removed by excisional biopsy under local anesthesia and sent to the Department of Pathology of Cumhuriyet University Faculty of Medicine for histopathological examination. Histopathological examination revealed uniform thickening of the epithelium and koilocytic squamous epithelial cells in the cytoplasm (Figure 2), which led to the diagnosis of FEH. The patient received regular cryosurgery treatment with the Cortex CryoPro device at the Department of Dermatology of Cumhuriyet University Faculty of Medicine every 15 days for 3 months. In the treatment, liquid nitrogen gas compressed at -196 degrees was used. Anesthesia was not applied to the relevant areas before the operation, no medication was given after the operation, and the lesions were left to heal spontaneously. The patient could not return to the clinic for follow-up at 1 year but she reported a marked improvement in the lesions, which was evident in the intraoral photographs that she had taken (Figure 3).

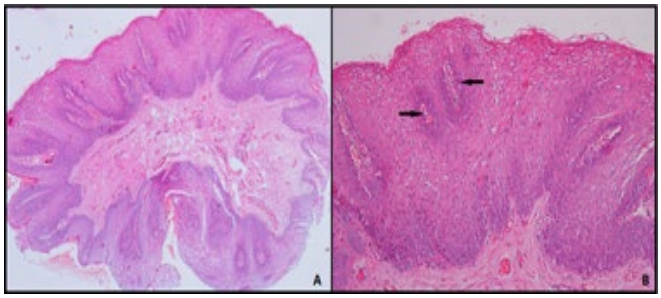

Figure 2.

(A) Uniform thickening of the surface epithelium, (B) Koilocytes (arrows) in the surface epithelium.

Figure 3.

Final appearance of the oral mucosa in the first year after treatment.

DISCUSSION

FEH is a rare epithelial proliferation induced by HPV ([10]). The oral cavity may host a number of HPV-related lesions, some of which are benign both microscopically and behaviorally. These include FEH, condyloma acuminatum, verruca vulgaris and squamous papilloma. Human-to-human transmission has been considered as the primary mode of contact in FEH etiology. This may explain familial clustering of FEH cases ([11]). In addition, malnutrition, poor hygiene and general health status may be associated with the disease ([10]). There are also studies reporting that environmental factors such as poor sanitation have an etiological role in FEH ([12]). The patient described in this case was from a family of moderate income.

FEH can be diagnosed on the basis of clinical, histopathological findings and polymerase chain reaction (PCR) method ([13]). In the oral mucosa, HPV infections may present with various clinical manifestations. HPV types 6 and 11 are implicated in the etiopathogenesis of oral verrucas and oral florid papillomatosis, and HPV types 13 and 32 in the etiopathogenesis of FEH ([13]). In a study suggesting that HPV 24 is the etiological factor in long-standing FEH cases, malignant transformation was reported, although the accuracy of the clinical diagnosis is doubtful ([14]). In the present case, the diagnosis was based on clinical and histopathological findings. Following the biopsy or excision of the lesion, a PCR analysis could not be performed to determine the exact HPV type that caused the pathology.

It is difficult to predict the long-term behavior of FEH lesions. Lesions may regress over the years and therefore, aggressive treatment is not required initially. Treatment can be considered in the case of increased lesion size and associated aesthetic concerns or pain or bleeding due to trauma ([15]). In the current case, treatment was provided due to the patient's aesthetic concerns.

The differential diagnosis of FEH includes verruca vulgaris, condyloma acuminatum, papilloma, irritation fibroma, juvenile papillomatosis, verruciform xanthoma and syndromes such as Cowden's syndrome, multiple endocrine neoplasia, neurofibromatosis, Gorlin-Goltz syndrome and tuberous sclerosis ([16]). Verruca vulgaris and papilloma are usually small proliferations of wart-like growths. Verruciform xanthoma is usually a pink colored lesion and occurs in areas exposed to irritation or trauma. Multiple endocrine neoplasia syndrome type IIb is characterized by sessile lesions on the tongue. The patient may have a marfanoid facial appearance, and there is an increased risk of pheochromocytoma and thyroid medullary carcinoma. Neurofibromatosis may be associated with epilepsy, cutaneous pigmentation and other neurological symptoms. Tuberous sclerosis might be associated with subungual fibroma, enamel defects, cutaneous hypopigmentation, epilepsy or mental retardation. Cowden's syndrome is typically associated with multiple hamartomas and patients carry an increased risk of developing cancers (especially of breast and thyroid). The Gorlin-Goltz syndrome is characterized by keratocyst formation, multiple nevoid basal cell carcinoma and skeletal abnormalities. All of these diagnoses can be ruled out by clinical examination and accurate history. Condyloma acuminatum and irritation fibroma are the most confusing lesions. In condyloma acuminatum, clustered papules develop on the ventral surface of the tongue and the floor of the mouth because of orogenital contact with an infected partner. As observed in this case, FEH lesions tend to be more numerous and flatter and the lesions are usually located in the lip and buccal mucosa. Irritation fibroma is distinguished from FEH by its pale appearance, enlargement over time and irritation on its surface ([14]).

Different methods can be used for the treatment of lesions. Surgical excision is a widely used method in FEH treatment. However, it has significant disadvantages including postoperative pain, scar and bleeding. CO_2 laser treatment can be preferred because of its hemostatic action and recovery without scar. Topical or systemic administration of interferon has been reported to be beneficial, especially in cases of extensive involvement ([3]).

Cryosurgery treatment is based on the principle of causing cell death via vasoconstriction, preventing blood flow to the main lesion. Two different methods are used in cryosurgery including open and closed techniques. In the open technique, liquid nitrogen is applied with cotton pellets, whereas in the closed technique, liquid nitrogen is applied directly to the lesion surface using various probes. The closed method provides a deeper surgery. In cryosurgery, tissues begin to freeze at -2.2° C and die at -20° C. During post-procedure recovery, more mature collagen is formed, which ensures less scar formation compared to other methods. In addition, the method is highly preferable since it does not require anesthesia, and patients do not experience bleeding or pain during or after the procedure ([17]). Moreover, it is easy to apply and has a low cost. The procedure does not cause dysfunction, aesthetic problems and scar, and provides a rapid recovery. Due to these advantages, cryosurgery is widely used in the treatment of a variety of pathological masses in general medicine and dental surgery. In dentistry, it is used for the treatment of soft and hard tissue tumors, local aggressive bone lesions, leukoplakia, gingivectomy, vascular anomalies, prosthetic hyperplasia, palatal hyperplasia, fibrous epulis, viral warts, mucous cysts and polyps ([18]). In the present case, closed cryosurgery method was used for the treatment of all lesions in the oral mucosa because of its advantages, and a significant improvement was achieved in the patient who received cryotherapy at 15-day intervals for 3 months. The patient is still being followed with no recurrence observed over 2 years after treatment.

CONCLUSION

After making a clinical and histopathological diagnosis of the rare FEH disease, the lesions may regress or remain same in size. The cryosurgery method, which can be applied for reasons such as aesthetic complaints and biting trauma, provides a rapid improvement and increases the patient's quality of life and motivation.

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