

***ORAL SQUAMOUS PAPILLOMA IN SIBLINGS: CASE REPORT
KARDEŞLERDE GÖRÜLEN ORAL SKUAMÖZ PAPILLOM: VAKA RAPORU**

Zeynep Burçin GÖNEN¹, Cihan TOPAN², Alper ALKAN³

¹Erciyes University, Genom and Stem Cell Centre, Kayseri

²Aksaray Oral and Dental Health Hospital, Private Practitioner, Aksaray

³Alper Alkan³ Bezmialem University, Department of Oral and Maxillofacial Surgery, Faculty of Dentistry, Istanbul

ABSTRACT

Oral squamous papilloma (OSP) is a benign growth of epithelium and minor amounts of supporting connective tissue. Its appearance varies between pink and white exophytic granular or cauliflower-like surface alterations. Although the etiology of OSP is not clear, human papilloma virus, which is a member of papova virus family, is assumed to be the etiologic agent of OSP. This article presents two cases who are siblings and have multiple papillary and verrucous exophytic lesions in different regions of the mouth. Based on clinical and histopathological evaluations the diagnosis was concluded as OSP. After a follow-up of one year the OSP lesions were regressed in both children idiopathically without any treatment.

The aim of this study is to describe the clinical and histopathologic features of multiple OSP lesions and to provide information on treatment methods.

ÖZ

Oral skuamöz papilloma (OSP), epitel ve az miktarda bağ dokusunun iyi huylu oluşumudur. Lezyonların rengi pembeden beyaza, şekli yüzeyden kabarık taneli veya karnabahar görümlü olarak farklılık gösterir. OSP'nin etyolojisi net olmamakla birlikte, papova virüs ailesinin bir üyesi olan insan papilloma virüsü lezyonların oluşumundan sorumlu tutulmaktadır.

Bu makalede, ağız içlerinin farklı bölgelerinde yüzeyden kabarık tomurcuk ve siğil benzeri lezyonları olan iki kardeşin olgu sunumu yapılmıştır. Lezyonlar klinik ve histopatolojik olarak değerlendirilmiş ve OSP tanısı konulmuştur. Bir yıllık takip sonrası OSP lezyonlarının her iki kardeşte de herhangi bir tedaviye gerek kalmadan kendiliğinden iyileştiği görülmüştür.

Bu çalışmanın amacı, çoklu OSP lezyonlarının klinik, histopatolojik özelliklerini tanımlamak ve tedavi yöntemleri hakkında bilgi vermektir.

Keywords: epithelial tissue, human papilloma virus; oral squamous papilloma

Anahtar kelimeler: epitel doku, insan papilloma virüsü; oral skuamoz papilloma

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Corresponding Author: Cihan Topan, DDs, PhD, Private Practitioner, Aksaray Oral and Dental Health Hospital Aksaray, Turkey
68200
E-mail: cihantopan@hotmail.com
Phone : +90 555 645 65 87
Fax : +90 382 213 51 48

INTRODUCTION

Oral squamous papilloma (OSP) is the benign growth of epithelium and minor amounts of supporting connective tissue resulting in papillary or verrucous exophytic mass. Its appearance changes pink to white exophytic granular or cauliflower-like surface alterations. The lesion generally measure not larger than one cm in greatest dimension but sometimes large lesions may be observed (1). Oral and oropharyngeal squamous cell papillomas occur mostly between three and five decades of life, however they may also emerged below the first decade. These lesions mainly localized in the areas of tongue and soft palate but any surface of the oral cavity can be affected (2). As an oral lesion, it may increase doubt due to its clinic appearance, which can imitate exophytic carcinoma, verrucous carcinoma, verruca vulgaris, multifocal epithelial hyperplasia or condyloma acuminatum. Although the etiology of OSP is not clear, human papilloma virus (HPV), which is a member of papova virus family, is assumed the etiologic agent of OSP (2,3). The aim of this study is to describe the clinical and histopathological features of multiple OSP lesions and present the regressed lesions in siblings.

CASE REPORT

We report the cases of two siblings, 15-years old female (patient 1) and 6-years old male (patient 2), born in Turkey. Lesions of female presented with multiply, well-defined, soft, asymptomatic growth on mucosa region of both side of the cheeks at the junction line of the upper lip and labial mucosal region. Lesions on the upper re-

gion of the tongue are seen multiply, whitish-pink appearance, verrucous growth. Lesions on the junction region of hard and soft palate presented with solitary, pedunculated, cauliflower appearance in shape (Fig 1). The patient noticed that lesion started from the left side of lower lip spread into the oral cavity three years previously. Medical, dental and personal histories were inconspicuous and no other abnormalities were noted on general physical examination.

Six years-old male, who is sibling of girl above, presented with multiply, painless growth on the left buccal mucosal region of the cheek and solitary, well-defined exophytic growth on the left side of lower lip and left commissura region (Fig 2). According to anamnesis which was taken from the mother; sister kissed her brother and she also used her brother's baby bottle to check the temperature of the milk with her lips and tongue.

Biopsy was performed on palatal region from female patient. Histopathological results were found to be consistent with oral squamous papilloma (Fig 3). After 1 year of follow-up the OSP lesions were regressed in both children idiopathically without any kind of treatment (Fig 4).

DISCUSSION

Oral squamous papillomas are classically divided into two groups: isolated- solitary and multiple-recurring. The former occur at any age but commonly between the ages of 30-50, found equally in men and women, while the latter is generally found in children (3,4). In present case report patients were under the age of 15-years-old

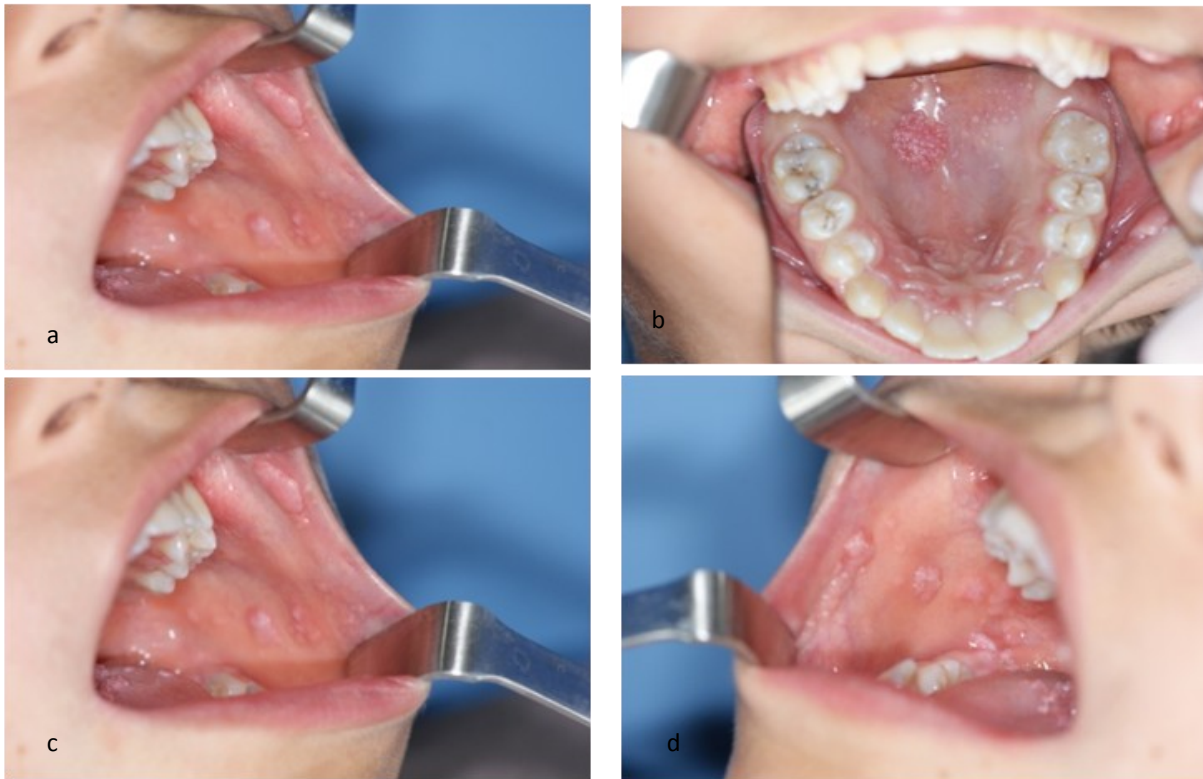


Figure 1. OSP lesions were detected on tongue (a), palate (b), and bilateral buccal mucosa (c,d) in female patient.



Figure 2. Well-defined, soft, asymptomatic growth on buccal mucosa in 6 year old male patient.

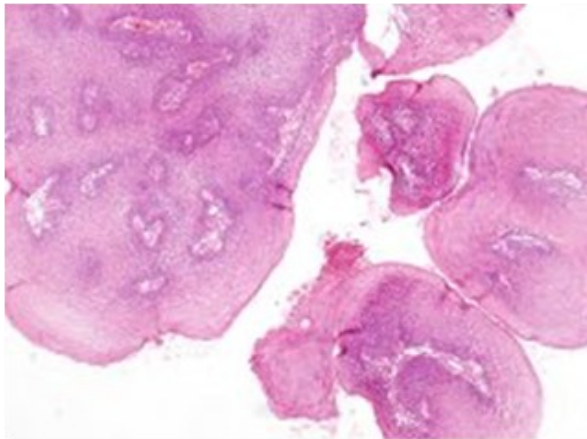


Figure 3. Biopsy was performed from female patient's palatal lesion. Irregular acanthosis and hyperkeratosis in stratified squamous epithelium. Basal membrane is intact. Chronic inflammatory cell infiltration in subepithelial area. There is no evidence to suggest a malignant lesion (Haematoxylin eosin, X100).



and lesions were multiple types in both patients, however no recurrence was observed.

OSP may be seen in any mucosal region in oral cavity including vermilion portion of the lips. Hard and soft palate and the uvula are the regions responsible for nearly one third of all regions (3). The lesions are usually asymptomatic and may be pedunculated or sessile in form. Pedunculated squamous papillomas generally occur in the areas of soft palate, tonsil or the epiglottis and can sometimes prove to be fatal. Non-keratinized lesions appear pink, whereas keratinized lesions are white in color (1,2). In our case report OSP lesions are detected on tongue, palate and buccal mucosal region of cheeks in patients. Lesions were asymptomatic in both patients consistent with literature.

All kinds of oral papillomas have viral origin is a debatable issue, but OSP have been shown to be associated with HPV subtype 6 and 11 (3,5). Clinically, lesions caused by HPV subtype 6 and 11 in oral cavity may be benign such as, focal epithelial hyperplasia, verruca vulgaris, condyloma acuminatum, common wart, oral leukoplakia. Oral squamous cell carcinoma, verrucous carcinoma and salivary gland adenocarcinoma are malign lesions caused by HPV subtype 16 and 18 (5,6). HPV 16 and 18 subtypes are also responsible for 90% of anogenital cancers (7). The viral presence should be detected with by using light microscopy, electron microscopy and molecular methods including amplified and non-amplified techniques (5). In histological examination koilocytic cell may be seen which is thought to be the determinative of a virally-affected area (3). In our histopathologic examination, koilocytosis is seen in the upper layers of the epithelium and there is no evidence to suggest a malignant lesion. Our diagnoses were based on clinical and histopathological evaluations.

HPV infects both sexual and non-sexual way. Possible

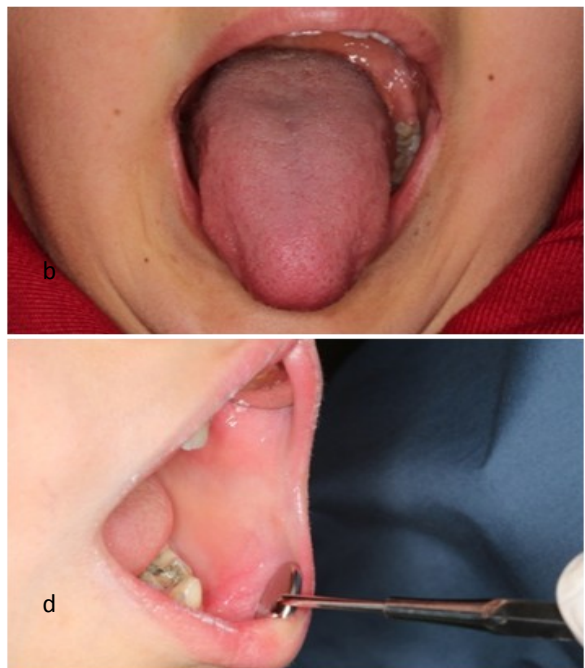


Figure 4. OSP lesion was regressed also in both female (a,b,c) and male and male (d) patient idiopathically.

non-sexual transmission routes are vertical or horizontal transmission and autoinoculation. HPV transmission usually occurs in horizontal way and virus is highly resistant to fatal effects like heat and dehydration (5,8). Although the route of transmission of the virus is unknown for oral lesions, direct contact would be favored as in present siblings.

Both medical and surgical techniques are used for the treatment of OSP. Lesions are treated surgically either routine excision or laser ablation. Electrocautery and cryosurgery may be considered as other treatment options. In medical treatment option agents like podophylin, isotretinoin, interferon alpha-2a, trikloseric acid, 5-FU, simethicone are used. Recurrence is uncommon, except the HIV (+) infected- patients (3). On the other hand limited surgical excisions end up with frequent recurrence owing to presence of HPV in latent form around the lesions. For this reason, surgical excisions are used with other treatment modalities like interferons (9). Vaccination may also be considered as an effective way of protection against HPV infections (1,10). OSP has a benign course with spontaneous regression; however have been associated with progression or persistence for years. In these present cases, after 1 year of follow-up the OSP lesions were regressed in both children without any kind of treatment.

As a conclusion, the clinical appearance of OSP resembles other benign and malignant oral lesions and differential diagnosis must be made with these lesions. It should be keep in mind that spontaneous regression may observe in HPV lesions. The follow-up the OSP lesions may be suggested for idiopathic regression for pediatric patients. When pediatric patient has OSP lesions, parents should be informed about direct transmission for preventing other family members.

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