Selcuk Dental Journal 10/1 (Nisan 2023): 94-96

Case Report / Olgu Sunumu

A Rare Case Report: Oral Focal Mucinosis

Nadir Görülen Bir Olgu:Oral Fokal Müsinöz

Kübra CERAN DEVECİ[α](ORCID-0000-0002-5962-7495), **Mehmet Reşat ÖZERCAN**[β](ORCID-0000-0003-3923-1642)

[α]Adıyaman Universty, Faculty of Dentistry, Departmen of Periodontology, Adıyaman, Turkey
[α]Adıyaman Üniversitesi, Diş Hekimliği Fakültesi, Periodontoloji Anabilim Dalı, Adıyaman, Türkiye
[β] Adıyaman Unıversıty Faculty Of Medıcıne Pathology Department, Adıyaman, Turkey
[β]Adıyaman Üniversitesi Tıp Fakültesi Patoloji Anabilim Dalı, Adıyaman, Türkiye

ABSTRACT

Oral focal mucinosis (OFM); It is a rare soft tissue lesion of unknown etiology. It was first described by Tomich in 1974. It usually occurs as a localized, asymptomatic pedunculated or sessile growth on the gingiva or hard palate. In the intraoral examination of a 14-year-old systemically healthy female patient who applied to our clinic with the complaint of gingival enlargement in the maxillary palate region, a well-circumscribed mass of approximately 2 cm in diameter, covered with normal mucosa, extending from the gingival to the hard palate, in the palatal region of the right first premolar and first molar teeth. The patient stated that he noticed the mass 3-4 years ago and did not experience any pain. The lesion was taken with excisional biopsy under local anesthesia and sent to the pathology department. The patient was diagnosed with OFM as a result of pathological evaluation. No recurrence after 1 year.

Keywords: Connective tissue diseases; oral health; oral pathology

ÖZ

Oral fokal müsinoz (OFM); etiyolojisi bilinmeyen, nadir görülen bir yumuşak doku lezyonudur. İlk kez 1974'te Tomich tarafından tanımlanmıştır. Genellikle dişeti veya sert damakta lokalize, asemptomatik saplı veya sapsız büyüme olarak ortaya çıkar. Kliniğimize üst çene damak bölgesinde bulunan dişeti büyümesi şikâyetiyle başvuran 14 yaşındaki sistemik olarak sağlıklı kadın hastanın ağız içi muayenesinde, sağ birinci premolar ve birinci molar dişlerinin palatinal bölgesinde, dişetinden sert damağa uzanan, normal mukoza ile kaplı, yaklaşık 2 cm çapında, iyi sınırlı bir kitle saptandı. Hasta kitleyi 3-4 yıl önce fark ettiğini ve herhangi bir ağrı yaşamadığı belirtti. Lezyon lokal anestezi altında eksizyonel biyopsi ile alındı ve patoloji bölümüne gönderildi. Hastaya patolojik değerlendirme sonucu ile OFM tanısı konuldu. 1 yılın sonunda nüks görülmedi.

Anahtar Kelimeler: Ağız sağlığı; bağ dokusu hastalıkları;oral patoloji

Introduction

Oral focal mucinosis (OFM) is a rare lesion, accepted as counterpart of cutaneous focal mucinosis on oral mucosa. Its' etiology has not known totally, it is though that it results from excessive hyaluronic acid production by fibroblasts.[1] It was first described by Tomich in 1974.[2] It is clinically seen as a round or oval shaped, painless, sessile or pedunculated lesion with the same color as the surrounding mucosa. It usually seen in adults in the fourth and fifth decades of life, but can also occur in children. The gingiva or hard palate is the most affected areas. The cases in other are such as buccal mucosa, tongue and lips have been reported in the literature. Since there is no pathognomonic finding in the examination, the final diagnosis is established by histological evaluation.[3,4] Histopathologically, a lesion consisting of a myxomatous connective tissue with clear borders, containing fusiform, oval or stellate fibroblasts is observed under the epithelium. A few capillary vessels can be observed in the lesion. Its treatment is surgical excision and recurrence is not usually seen.5As far as we are concerned, OFM cases of about 120 around the world have been reported so far.[3] We also presented our rare OFM case for the purpose of contributing to the literature in this study.

Case Report

A 14-year-old female patient with the complaint of gingival hyperplasia in the maxilla and palate presented to Adıyaman University Faculty of Dentistry Periodontology Clinic. Anamnesis taken from the patient it was learned that she had recognized the mass 3-4 years ago and had not experienced any pain. In the intraoral examination, a well-circumscribed mass of approximately 2 cm in diameter, covered with normal mucosa, reaching from the gingiva to the hard palate, was found in the palatal region of the right first premolar and first molar teeth (**Figure 1**).

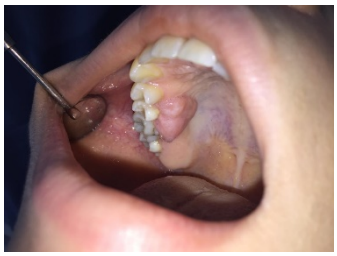


Figure 1. Intraoral view of the lesion

The lesion was firm and painless on palpation. Radiographic examination was normal. The lesion was taken by excisional biopsy under local anesthesia and sent to the pathology department. In the macroscopic examination, it was observed as a nodular mass of 1.8x1.1x0.3 cm, covered with mucosa, and of elastic consistency. In the microscopic examination, loose myxomatous connective tissue areas, surrounded by dense collagenized areas in the periphery, covered with stratified squamous epithelium, were seen in the sections. The cells were rare, fusiform and stellate in this areas **(Figure 2a, b).**

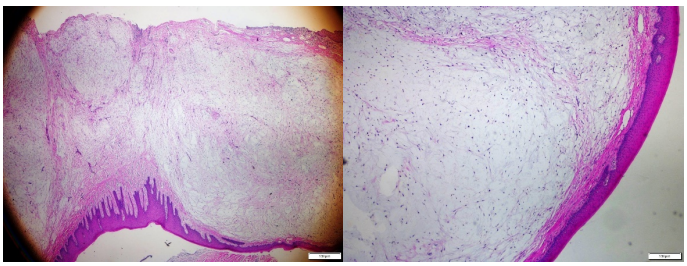


Figure 2. a) Hematoxylin eosin (X40) **b)** Stratified squamous epithelium on the surface, loose myxomatous stroma under the epithelium (Hematoxylin eosin X100)

Gönderilme Tarihi/Received: 20 Ekim, 2022
Kabul Tarihi/Accepted: 28 Ocak, 2022
Yayınlanma Tarihi/Published: 27 Nisan, 2023
Atıf Bilgisi/Cite this article as: Ceran Deveci K, Özercan MR. A Rare Case Report: Oral Focal Mucinosis. Selcuk Dent J 2023;10(1):94-96 Doi: 10.15311/ selcukdentj.1192103

Sorumlu yazar/Corresponding Outhor: Kübra Ceran Deveci
E-mail: k_crn@hotmail.com
Doi: 10.15311/ selcukdentj.1192103

Mucinous areas were PAS (Periodic Acid-Schiff) dye gave a negative reaction, while it showed a positive reaction with alcian blue (**Figure 3**). OFM diagnosis was established to patient in consequence of pathologic evaluation. The patient was followed up at regular intervals. No recurrence was observed at the end of 1 year.(**Figure 4**).

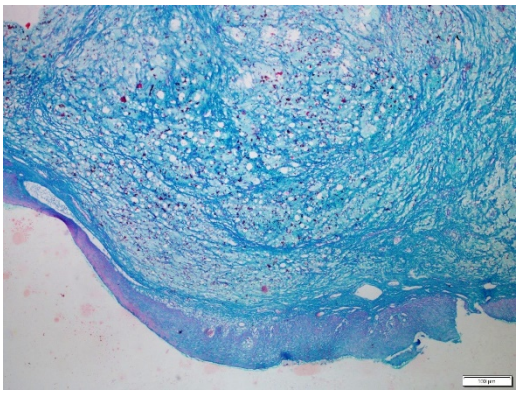
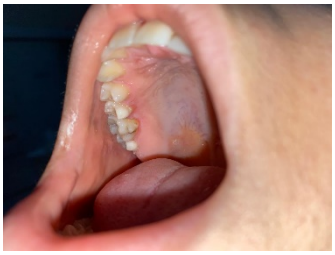


Figure 3. Myxomatous stroma, positive alcian blue stain and negative PAS (X100)



Figüre 4. 1-year follow-up

Discussion

OFM is a mucosal disease that is rare and has unknown etiology. Although the pathophysiology is not clearly understood, it has been suggested by Tomich 2 that it occurs due to the excessive hyaluronic acid production by fibroblasts and its positive staining with alcian blue supports this view.[6] It is commonly observed in the fourth and fifth decades of life and it is more common tin females than males by a ratio of 2:1.[3] It was stated that the number of individuals under the age of 18 who were established OFM diagnosis was less than 10 in a study published by Cameron et al in 2020.[4] Our case was a 14-year-old female patient.

Clinically, neoplastic and inflammatory lesions can be confused such as fibrous hyperplasia, squamous papilloma, giant cell fibroma, peripheral giant cell granuloma, peripheral ossifying fibroma, peripheral odontogenic fibroma and pyogenic granuloma. OFM occurs as pedunculated or sessile, painless, nodular mass the same color as normal mucosa.[5,7] Ulceration is not seen on its surface however it has been reported in the literature in cases where ulceration is seen.[8] Its size changes from a few mm to 2 cm.[7] In our case, the largest diameter was found to be 1.8 cm.

The histopathologic image of OFM is similar to lesions in myxomatosis features such as neurofibroma, nerve sheath myxoma, mucocele, odontogenic myxoma, and soft tissue myxoma. Intraoral soft tissue myxoma is a rare mesenchymal tumor and it is composed of stellate or fusiform cells with a reticular fiber in the loose mucoid stroma. It is encapsulated and may invade surrounding tissues. Oral focal mucinosis is distinguished from this lesion and other myxomatosis lesions because of its clear borders and absence of reticulin fiber.[9,10] Another lesion that should be considered in the differential diagnosis of OFM is odontogenic myxoma. Odontogenic myxoma is a rare nonmalignant mesenchymal odontogenic tumor localized in the oral cavity, usually in the mandible. Unlike OFM, it has a locally aggressive feature and a high recurrence rate.[11] The fact that the mucocele contains granulation tissue is its distinguishing finding from OFM.[4]

The standard treatment for OFM is surgical excision and recurrence is not usually seen. It is reported that recurrence was seen in only seen two cases in the literature.[12] No recurrence was observed in the follow-up of the patient.

Conclusion

OFM, in the literature, is a nonmalignant oral soft tissue lesion reported that is rarely seen especially under the age of 18. It is clinically similar to other oral mucosal lesions, so histological examination is required for definitive diagnosis. It is treated with surgical excision and it usually is not observed recurrence. Clinically, due to the absence of pathognomonic features, it is a pathology that should be kept in mind by dentists and pathologists in the differential diagnosis of oral lesions.

Değerlendirme / Peer-Review

İki Dış Hakem / Çift Taraflı Körleme

Etik Beyan / Ethical statement

Bu olgu sunumu Türk Periodontoloji Derneği 51. Uluslararası Bilimsel Kongresi 29. Bilimsel Sempozyumunda (Kasım 2022) sözlü bildiri olarak sunulmuştur.

Bu çalışmanın hazırlanma sürecinde bilimsel ve etik ilkelere uyulduğu beyan edilmiş ve yararlanılan tüm çalışmalar kaynakçada belirtilmiştir.

This case report was presented as an oral presentation at the 29th Scientific Symposium of the 51st International Scientific Congress of the Turkish Society of Periodontology (November 2022).

It is declared that during the preparation process of this study, scientific and ethical principles were followed and all the studies benefited are stated in the bibliography.

Benzerlik Taraması / Similarity scan

Yapıldı - ithenticate

Etik Bildirim / Ethical statement

ethic.selcukdentaljournal@hotmail.com

Çıkar Çatışması / Conflict of interest

Çıkar çatışması beyan edilmemiştir.

Finansman / Grant Support

Yazarlar bu çalışma için finansal destek almadığını beyan etmiştir. | The authors declared that this study has received no financial support.

Çıkar Çatışması / Conflict of Interest

Yazarlar çıkar çatışması bildirmemiştir. | The authors have no conflict of interest to declare.

Yazar Katkıları / Author Contributions

Çalışmanın Tasarlanması | Design of Study: KCD (%60), MRÖ (%40)

Veri Toplanması | Data Acquisition: KCD (%40), MRÖ (%60)

Veri Analizi | Data Analysis: KCD (%35), MRÖ (%65)

Makalenin Yazımı | Writing up: KCD (%60), MRÖ (%40)

Makale Gönderimi ve Revizyonu | Submission and Revision: KCD (%65), MRÖ (%35)

REFERENCES / KAYNAKLAR

1. Nilesh, K., Kothi, HS., Patil R., Pramod RC.Oral focal mucinosis of posterior maxilla. Journal of Oral and Maxillofacial Pathology: 2017;21(2):273.
2. Tomich CE. Oral focal mucinosis: a clinicopathologic and histochemical study of eight cases. Oral Surg Oral Med Oral Pathol 1974;38:714–24.
3. Silva Cunha, JL., Leite AA., de Castro Abrantes T., Vervloet LP., de Lima Morais, TM., de Oliveira Paiva Neto, G., ... & Soares CD.Oral focal mucinosis: A multi-institutional study and literature review. Journal of Cutaneous Pathology, 2021;48(1);24-33.
4. Cameron A., Webster JEN., Wicks E., Colbert SD.Oral focal mucinosis of the palate: a rare disease entity. BMJ Case Reports 2020;13(3):e230233.
5. Lee JG., Allen G., Moore, L., Gue S.Oral focal mucinosis in an adolescent: a case report. Australian dental journal.2012;57(1):90-2.
6. Buchner A, Merrell PW, Leider AS, Hansen LS. Oral focal mucinosis. Int J Oral Maxillofac Surg. 1990;19(6):337-40.
7. Talacko AA., Lacy MF., Besly WJ., Aldred MJ.Oral focal mucinosis: report of two cases with ulceration. Pathology.2004;36(6):582-3.
8. Madhusudhan AS, Nagarajappa D, Manjunatha BS, Saawarn Swati, Charan Babu HS. Oral focal mucinosis: report of two cases. Rev Odonto Cienc 2010;25:310–3.
9. Higuchi Y., Tsushima F., Sumikura K., Sato Y., Harada H., Kayamori K., Ikeda, T. Diagnosis and treatment of oral focal mucinosis: a case series. Journal of Medical Case Reports.2019;13(1);108.
10. Tekkesın MS, Yilmaz M, Olgaç V. Oral focal mucinosis: a case report and review of the literature. Turk Patoloji Derg. 2013;29(3):235-7.
11. MacDonald-Jankowski DS, Yeung R, Lee KM, Li TK. Odontogenic myxomas in the Hong Kong Chinese; clinico-radiological presentation and systematic review. Dentomaxillofac Radiol 2002;32:71-83.
12. Bharti V, Singh J. Oral focal mucinosis of palatal mucosa: A rare case report. Contemp Clin Dent. 2012;3(2):214-8.