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Malign or Benign Lesion? Isolated Plasma Cell Mucositis of the Lip: A Rare Case

Malign veya Benign Lezyon? Dudakta İzole Plazma Hücreli Mukozit: Nadir Bir Olgu

Uğur ERGÜN 

Medicinae Doctor, Department of Internal Medicine, Balıkesir Atatürk City Hospital, Balıkesir, Türkiye.

Sorumlu Yazar | Correspondence Author

Uğur ERGÜN

mdbalkes10@gmail.com

Yazışma Adresi | Address for Correspondence: Gaziosmanpaşa, 209 Street, No:26 Altıeylül, Balıkesir, Türkiye.

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Şikayetler: hmj@hitit.edu.tr

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Malign or Benign Lesion? Isolated Plasma Cell Mucositis of the Lip: A Rare Case

Abstract

Background: Plasma cell mucositis is a rare, benign, chronic inflammatory disease that is typically localized in the mucosa of the oral cavity.

Case Report: In this case report, a 28-year-old woman presented with a painless swelling in the corner of her lip for three months, and histopathological examination revealed a diagnosis of plasma cell mucositis. The lesion's isolation to lips corner suggests a rare variant of plasma cell mucositis. In the differential diagnosis, malignant plasma cell disorders, infections, and autoimmune diseases were considered. However, laboratory tests and imaging methods ruled out underlying systemic plasma cell dyscrasia. Clinical improvement was achieved after surgical excision, and no recurrence was observed during follow-up.

Conclusion: This case report aims to raise awareness that the rare localization of plasma cell mucositis on the lip should not be confused with malignancy.

Keywords: Plasma cell mucositis, lip lesion, differential diagnosis, malignancy.

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Öz

Giriş: Plazma hücreli mukozit, genellikle ağız boşluğu mukozasında lokalize olan, nadir görülen, benign ve kronik inflamatuvar bir hastalıktır.

Olgu Sunumu: Bu olgu sunumunda, 28 yaşında kadın hasta, dudak köşesinde üç aydır devam eden ağrısız şişlik ile başvurmuş olup, yapılan histopatolojik inceleme sonucunda plazma hücreli mukozit tanısı konulmuştur. Lezyonun dudak köşesi ile sınırlı olması, plazma hücreli mukozitin nadir bir varyantını düşündürmektedir. Ayırıcı tanıda malign plazma hücre hastalıkları, enfeksiyonlar ve otoimmün hastalıklar değerlendirilmiştir. Ancak laboratuvar testleri ve görüntüleme yöntemleri altta yatan sistemik plazma hücre diskrazisini dışlamıştır. Cerrahi eksizyon sonrası klinik iyileşme sağlanmış ve takip süresince nüks izlenmemiştir.

Sonuç: Bu olgu sunumu, plazma hücreli mukozitin dudakta nadir görülen lokalizasyonunun malignite ile karıştırılmaması gerektiğine dikkat çekmeyi amaçlamaktadır.

Anahtar Kelimeler: Plazma hücreli mukozit, dudak lezyonu, ayırıcı tanı, malignite

Introduction

Plasma cell mucositis (PCM) is a rare, benign, chronic inflammatory disease. These lesions, typically localized in the mucosa of the oral cavity and pharynx, are characterized by dense plasma cell infiltration (Solomon & Schow, 1988; Solomon, Wein, & Rosenwald, 2008). Clinically, lesions may present as painless swelling, erythema, edema, or mucosal thickening (Segura & Pujol, 2008). The gold standard for diagnosis is histopathological examination, and in the differential diagnosis, malignant plasma cell disorders (such as multiple myeloma or plasmacytoma), infectious agents, and autoimmune diseases should be considered (Woo, Lin, & Moutasim, 2014). The exact etiopathogenesis of PCM is not fully understood, but mechanisms such as localized antigenic stimuli or an exaggerated immune response are believed to play a role (Segura & Pujol, 2008). A limited number of cases have been reported in the literature, and the clinical course is generally benign. However, due to its rarity, delays in diagnosis may occur, and unnecessary invasive procedures or further tests to support malignancy suspicion may be performed (White, Olsen, & Banks, 1986).

This case report presents the clinical evaluation, differential diagnosis process, and follow-up results of a young female patient who presented with a palpable lesion on the lip mucosa for three months and was diagnosed with PCM based on histopathological examination.

Case Report

A 28-year-old female patient presented to a tertiary healthcare facility with a painless swelling in the left lower corner of the lip that had been present for approximately three months. The patient had no known systemic diseases, chronic medication use, or history of smoking or alcohol consumption. There was no family history of malignancy. Upon initial presentation, the patient was evaluated in the dermatology clinic and started on topical steroid therapy, but no clinical response was observed. Due to persistent swelling, the patient voluntarily sought further evaluation at the otolaryngology clinic, where a well-defined, soft, approximately 2 cm lesion was noted at the left lower lip corner during physical examination. The lesion was excised via biopsy, and histopathological examination confirmed the diagnosis of PCM (Figure 1). The patient was referred to internal medicine and hematology clinics for evaluation of any underlying systemic plasma cell dyscrasia and was examined by our team. Laboratory tests, including complete blood count, biochemical function tests, peripheral smear, serum immunoglobulin levels (IgA, IgG, IgM), serum protein electrophoresis (Figure 2), immunofixation electrophoresis, free and total light chain analyses (kappa, lambda), and beta-2 microglobulin levels, were all within normal limits (Tables 1–2). In addition, C-reactive protein (CRP) levels measured during the three-month follow-up period were within normal limits. An oncological screening using F-18 FDG PET/CT imaging revealed no pathological focal areas with abnormal metabolic activity. No clinical, laboratory, or imaging findings suggestive of systemic plasma cell dyscrasia were observed, and the condition was

considered a benign case of plasma cell mucositis. As the patient remained asymptomatic, no additional treatment was planned, and follow-up was scheduled at six-month intervals in the outpatient clinic.

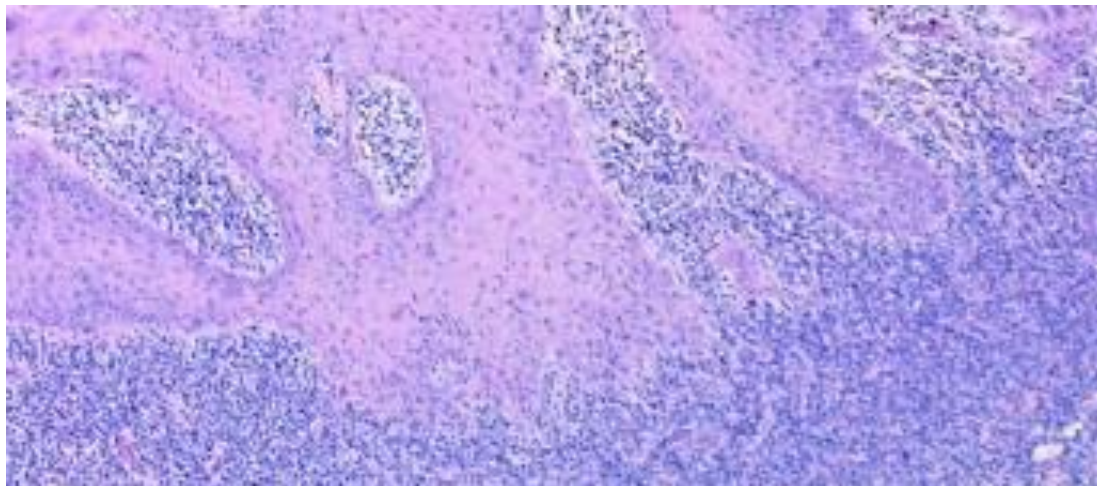


Figure 1. Ulceration in the oral mucosa and dense plasma cell infiltration in the underlying connective tissue (Hematoxylin and Eosin stain; magnification $\times 40$).

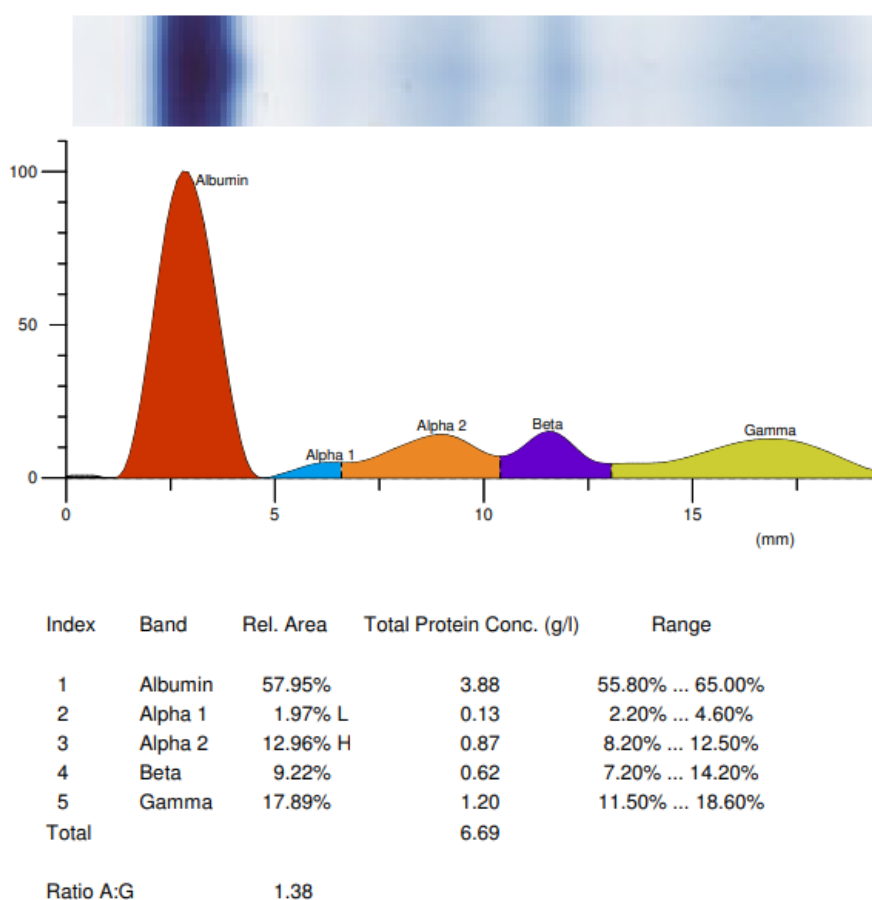


Figure 2. Serum Protein Electrophoresis

Table 1. Hemogram Parameter Results

Test Name	Result	State	Unit	Reference Range
WBC	8.26		$10^3/\mu\text{L}$	4.49 - 12.68
RBC	4.92		$10^6/\mu\text{L}$	3.92 - 5.08
HGB	14.8		g/dL	11.9 - 14.6
HCT	43.8		%	36.6 - 44.0
MCV	89.0		fL	82.9 - 98.0
MCH	30.1		pg	27.0 - 32.3
MCHC	33.8		g/dL	31.8 - 34.7
PLT	190		$10^3/\mu\text{L}$	150 - 450
PCT	0.20		%	0.18 - 0.39
PDW	13.0		fL	9.9 - 15.4
MPV	10.4		fL	9.1 - 11.9
NEU#	4.33		$10^3/\mu\text{L}$	2.1 - 8.89
LYM#	3.12		$10^3/\mu\text{L}$	1.26 - 3.35
MONO#	0.67		$10^3/\mu\text{L}$	0.25 - 0.84
EOS#	0.10		$10^3/\mu\text{L}$	0.01 - 0.4
BASO#	0.04		$10^3/\mu\text{L}$	0.01 - 0.07
NEU%	52.4		%	42.90 - 74.30
LYM%	37.8		%	18.3 - 45.7
MONO%	8.1		%	4.2 - 11.8
EOS%	1.2		%	0.20 - 5.30

Table 2 Monoclonal Gammopathy/Plasma Cell Dyscrasia Screening Panel

Test Name	Result	Unit	Reference Range
Beta-2 Microglobulin	1.5	mg/L	0.9 - 2.7
Immunofixation Electrophoresis	.		
Kappa Light Chain, Free	15.7	mg/L	0.6 - 22.4
Kappa Light Chain, Total	3.1	g/L	1.7 - 3.7
Lambda Light Chain, Free	19.2	mg/L	8.3 - 27
IgG	1270	mg/dL	700 - 1600
IgA	84.3	mg/dL	70 - 400
IgM	91	mg/dL	40 - 230

Discussion

PCM is a rare, benign inflammatory disease that typically occurs in the mucosa of the oral cavity. First described by Zoon (1952), this condition is most commonly observed in the gingiva, buccal mucosa, and tongue, and less frequently in the pharynx or lip (Solomon & Schow, 1988; Solomon et al., 2008). In the presented case, the lesion being localized exclusively to the lip corner aligns with the limited similar cases reported in the literature, which represent a rare variant of the disease (El-Khateeb & Zeitoun, 2012; Janam et al., 2012; Hanami et al., 2011).

The exact etiopathogenesis of PCM remains unclear. Chronic irritation, prosthetic use, contact allergens, and localized immune responses are thought to contribute to the development of the disease (Segura & Pujol, 2008). In addition, hypersensitivity reactions to certain dietary components and herbal substances have also been reported in the literature (Bali et al., 2012). However, some studies have also reported associations with autoimmune diseases (Rahal et al., 2015). In this case, no systemic disease or connective tissue pathology was observed.

Clinically, PCM can mimic hematologic malignancies and other neoplastic processes. The lesion is typically painless, nodular, or plaque-like, and may enlarge over time. Therefore, oral leukoplakia, erythroplakia, plasmacytoma, multiple myeloma, and infectious agents must be considered in the differential diagnosis (Woo et al., 2014; White et al., 1986). In addition, plasma cell gingivitis and related conditions should also be considered in the differential diagnosis (Konidena et al., 2014). Histopathological examination is required to confirm the diagnosis. In this case, an excisional biopsy was performed after failure of topical treatments, enabling an accurate diagnosis.

Histopathologically, PCM is characterized by dense and polyclonal plasma cell infiltration in the subepithelial area. Therefore, immunohistochemical studies and systemic evaluation are crucial to distinguish it from malignant plasma cell proliferations (Woo et al., 2014). The differential diagnosis

should especially exclude multiple myeloma, plasmacytoma, and other plasma cell dyscrasias. To achieve this, serum immunoglobulin levels (IgA, IgG, IgM), serum protein electrophoresis, free light chain analysis (kappa-lambda ratio), and systemic imaging methods are employed. In this case, all hematologic and biochemical parameters were within normal limits, and F-18 FDG PET/CT imaging showed no abnormal metabolic activity. These findings excluded the possibility of an underlying systemic plasma cell dyscrasia and supported the diagnosis of benign PCM.

Topical corticosteroids are commonly used as first-line treatment for oral cavity lesions. However, alternative therapies such as topical tacrolimus have also been reported to be effective in selected cases (Hanami et al., 2011). For limited, symptomatic lesions, surgical excision is both diagnostic and therapeutic (El-Khateeb & Zeitoun, 2012; Woo et al., 2014). In this case, significant clinical improvement was observed after surgical excision, and no recurrence was noted during follow-up. This supports the benign nature of PCM and indicates that, in suitable cases, surgical intervention may be sufficient for diagnosis and treatment.

This case report emphasizes the importance of the differential diagnosis of the rare lip variant of PCM and highlights the need to increase clinical awareness to prevent confusion with malignancy.

Conclusion

In conclusion, this case highlights a rare lip-localized variant of plasma cell mucositis, underlining the importance of careful differential diagnosis to distinguish it from malignant plasma cell disorders and other neoplastic conditions. The absence of systemic involvement, normal hematologic parameters, and negative PET/CT findings supported the benign nature of the lesion, while histopathological examination confirmed the diagnosis. Clinical improvement and the lack of recurrence following surgical excision further demonstrate that localized PCM can be effectively managed with surgical intervention when appropriate. This report emphasizes the need for heightened clinical awareness to avoid misdiagnosis and ensure proper treatment of this uncommon presentation.

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