A Rare Presentation of Chronic Exfoliative Cheilitis: Case Report

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ABSTRACT

BACKGROUND: Chronic exfoliative cheilitis is an inflammatory condition of the lips that involves exfoliation of the superficial layers of the lip with continuous peeling, ulceration, and exfoliation. Chronic exfoliative cheilitis can lead to social and psychosocial problems.

CASE PRESENTATION: A 73-year-old diabetic male presented with a lip lesion showing continuous ulceration, crusting, and peeling of the lower lip. It was a spontaneous complaint that had its onset 6 months ago and has constantly evolved. The medical history revealed controlled diabetes mellitus while the social and family histories showed no marked findings. After a careful history, clinical examination, and laboratory investigations with the exclusion of the possible etiologic factors of cheilitis, the patient was diagnosed with chronic exfoliative cheilitis with unknown etiology. Then, the patient was prescribed topical steroids, and he was advised for continuous follow-up visits.

CONCLUSION: Diagnosing chronic cheilitis is challenging and requires careful history, clinical examination, and laboratory investigation, with continuous follow-up to improve the patient's quality of life. Further research is needed to better understand and treat these cases.

KEYWORDS: Cheilitis, case reports, lip diseases, inflammation, steroid therapy

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Introduction

Chronic exfoliative cheilitis refers to the condition where one or both lips develop chronic inflammation. The affected lips can become so inflamed that they eventually form cracks. Over time, the condition can cause symptoms such as ulceration, tingling sensation, a red or purplish border on the lip, and peeling or crusting areas. Burning sensation in the affected area might also develop.^{1,2}

The exact cause of chronic cheilitis is currently unknown. It is generally thought to result from some type of everyday environmental exposure, a hypersensitivity to allergens found in food or drink, or the repeated application of commercial lip balm. In a few people, chapped lips or cheilitis can develop after they become sensitive to substances contained in dental care products or cocaine replacement. Chemotherapy, radiation therapy, and HIV infections have also been linked to the development of chronic cheilitis. Other causal factors might include nutrient deficiencies, climatic changes, or systemic disease. Due to the rarity of chronic cheilitis, it is commonly misdiagnosed as actinic or allergic cheilitis.^{3,4}

Cheilitis was categorized according to the etiology and duration of the disease into three categories. Reversible cheilitis including simplex, angular, infective, contact, eczematous, exfoliative, and drug-related cheilitis. Irreversible cheilitis includes actinic, granulomatous, glandular, and plasma cells. In

addition, there are cheilitis connected to dermatologic or systemic diseases such as lupus and lichen planus.⁵

We present the case of a 73-year-old male patient with a chronic lip lesion that initially posed diagnostic challenges and was characterized by recurrent, clinically relapsing, and severe symptoms.

Case Presentation

We report a clinical presentation of a 73-year-old Middle Eastern patient who presented to the Oral Medicine Clinic at the Faculty of Dentistry, Mansoura University, Egypt with a 6-month history of lip lesion. The patient's medical history showed controlled diabetes mellitus while the family and social histories are unremarkable, with no specific findings of note.

Examination of the lip revealed a persistent diffuse erosive lesion on the lower lip, measuring approximately 2×3 cm and extending from the mucocutaneous junction to the inner aspect of the lower labial mucosa. The lesion has irregular but well-demarcated margins that peels off and is characterized by an erythematous, edematous appearance with interspersed thickened white patches. Additional features include fissuring, bleeding, areas of necrosis, crusting, and continuous exfoliation, revealing a flaky texture.

Visual inspection reveals pronounced swelling of the lower lip, while palpation shows normal texture without induration or tenderness. The lower vermilion margin was associated with painful paresthesia and epidermal desquamation (Figure 1).

No vesicular or bullous lesions were observed. There was also no regional or generalized lymphadenopathy. In addition, no facial lesions or erosion were seen. A radiographic examination was requested to rule out any lingual or mandibular growth. The patient denied a history of smoking or chronic exposure to sunlight. He did not use any medication, lip care, or lip balm to alleviate these problems. In the intraoral examination, there were no signs of sharp fillings in the lower and upper jaws or injuries due to trauma. There were no other lesions in the oral cavity.



Figure 1. Clinical photograph showing a case of chronic exfoliative cheilitis with erythematous, edematous and white patches, bleeding, and necrotic appearance.

The blood count and serological tests of the patient were observed to be within the normal range in laboratory analyses (Table 1). These results indicated no significant abnormalities or underlying systemic conditions contributing to the patient's lip lesion.

We coordinated with the Oral Pathology Department to request an incisional biopsy to determine the diagnosis and prognosis as well as tailored treatment of the lesion. The histopathological report revealed the presence of a central area of ulceration and deep infiltration of polymorphs, pus cells, and lymphocytes in the labial submucosa and underlying muscle tissue. No signs of granuloma or malignancy were detected.

After rolling all other possible causes, the patient was diagnosed with chronic exfoliative cheilitis. He was instructed to use topical steroids (betamethasone valerate 0.1% topical ointment) twice daily. In addition, the patient was advised to avoid allergens and sun exposure. After 1 month of treatment, the patient showed complete healing of the lesion (Figure 2).

Discussion

Chronic exfoliative cheilitis is a rare inflammatory disease that may be challenging to diagnose due to its nonspecific clinical presentation and overlapping features with different sorts of cheilitis. The etiology remains mostly unknown, with possible contributing factors including chronic inflammation, hypersensitive reactions, or autoimmune diseases. The possible etiology should be reviewed. A smear can be carried out to help to prevent confusion with viral diseases. Histopathological examination of the affected mucosa and identification of the underlying cytological and architectural characteristics are other essential tools that lead to an early definitive diagnosis of

Table 1. The blood count and serological tests of the case.

TEST	RESULTS	NORMAL RANGE
Red blood cells count	$5.1 \times 10^6/\mu L$	$4.7-6.1 \times 10^6 / \mu L$
white blood cells count	7.2×10 ⁹ /L	4.0-11.0×10°/L
Hemoglobin (Hb)	14.1 g/dL	12.0–16.0 g/dL
Platelet count	235×10°/L	150-450 × 10°/L
Fasting blood sugar (FBS)	145 mg/dL	70-99 mg/dL (normal); 100-125 mg/dL (prediabetes); >126 mg/dL (diabetes)
Glycosylated Hemoglobin (HbA1c)	7.2%	<5.7% (normal); 5.7%-6.4% (prediabetes); ≥6.5% (diabetes)
Liver function tests		
Alanine Aminotransferase (ALT)	22 U/L	7-56 U/L
Aspartate Aminotransferase (AST)	18 U/L	10-40 U/L
Renal function tests		
Blood urea nitrogen (BUN)	14 mg/dL	6–20 mg/dL
Creatinine	0.9 mg/dL	0.6-1.3 mg/dL

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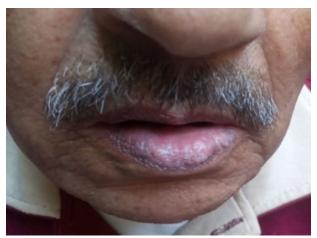


Figure 2. Clinical photograph showing complete healing of the lip lesion after 1 month of treatment.

the condition before any unnecessary investigations are performed. The clinical and histological features found in the reported case are consistent with the diagnosis of chronic exfoliative cheilitis.

In this case, the absence of any identifiable allergens or infection brought about a diagnosis of chronic exfoliative cheilitis of unknown etiology. The treatment approach targeted on lowering irritation, protecting the lips from further irritation, and preventing secondary infections. While corticosteroids are powerful in controlling the signs and symptoms, long-term management may require lifestyle modifications to prevent recurrence. The use of sunscreens, avoiding contact with allergens, and possible allergological and/or medicinal counseling are always useful measures. 4,6,8

The differential diagnosis of the condition includes contact cheilitis, recurrent herpes labialis, secondary syphilis, lip-licking dermatitis, factitial cheilitis and vitamin deficiency.^{5,6}

Due to the nature of the 6-month history of the lip lesion in our case, we differentiate it from the orofacial granulomatosis (OFG). It represents a rare inflammatory condition characterized by the presence of granulomatous inflammation in the soft tissues of the oral and maxillofacial region. In idiopathic OFG, the etiology is still unclear; however, a multifactorial origin has been proposed, involving genetic, immunological, and environmental factors. The most common presentation is the presence of chronic lip swelling; however, patients may show variable symptoms, occasionally developing systemic alterations. The disease can occur as Miescher's isolated granulomatous cheilitis or with other granulomatous diseases (Crohn's disease, sarcoidosis and Melkersson-Rosenthal syndrome). Melkersson-Rosenthal syndrome is characterized by tongue involvement, although only 1 or 2 symptoms usually appear (the complete triad of symptoms occurs in only 25% of patients). by granulomatous cheilitis, facial palsy and plicated tongue^{9,10}

Secondary orofacial granulomatosis: This type of OFG can be a manifestation of underlying systemic and regional granulomatous entities. Any diagnostically challenging papulonodular mucocutaneous disease should trigger a differential diagnosis workup including Crohn's disease, sarcoidosis, granuloma annulare, erythema nodosum, tuberculosis, leprosy, syphilis, leishmaniasis, cat-scratch disease, foreign body reaction, and, importantly, contact allergens. Secondary OFG does not require further diagnostic workup based solely on this presentation; however, discovery of the underlying etiology is crucial to guide further diagnostic or therapeutic management. 11-14

Stone et al¹⁵ reported a case of lip-lick cheilitis in an 8-year-old male who habitually licked his lips and presented with dry, flaky, and red skin bordering the outside of his lips. In addition, Mani and Shareef² reported a case of exfoliative cheilitis in an 18-year-old male with ulceration and crusting of the lips, which he had had for 1 year.

A dietary recommendation was given to avoid a mechanically aggressive diet, promoting cold, soft, and non-spiced foods. Additionally, he was advised to maintain good oral hygiene, lubricate the dental appliances on contact areas with the mucosa, avoid triggering foods, and use sunblock. We asked the patient to maintain regular follow-up visits. 16,17

The diagnosis of chronic cheilitis is very complex, leading to several episodes of inflammation and pain, worsening the patient's oral health and overall quality of life. With this work, we can verify that the specified treatment produced positive results in relieving symptoms, pointing to improvement in the adaptation and quality of life of the patient. The satisfactory results obtained in this case were undoubtedly due to a multi-disciplinary approach that made it possible to resolve the initially diagnosed condition. 3,4,6,18

Conclusion

This case highlights the importance of thorough medical evaluation and a multidisciplinary approach in managing chronic cheilitis. Although chronic exfoliative cheilitis is a very rare condition, recognizing its distinct clinical and histopathological functions is prime to differentiating it from other types of cheilitis. Early diagnosis and treatment can considerably improve patient outcomes and prevent complications such as secondary infection or scarring.

Author Contributions

Abdelrahman Eltoureini & Hamad Alharbi & Wafaa Saleh: Data curation, investigation and writing original draft. Wafaa Saleh: Data curation, investigation, supervising and editing original draft.

Informed Consent

Written informed consent was obtained from the patient

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