


Report of two cases of exfoliative cheilitis and brief literature review

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Abstract:

Exfoliative cheilitis is a rare skin disease that there is not consensus about its pathophysiology, clinical course and treatment. It can cause functional and aesthetic problems, being associated with psychiatric and/or emotional disorders. Our objective is to report two cases of patients with exfoliative cheilitis and provide records to the limited literature. Results obtained with Prednisone, lip balm, fluid extract of Chamomila recutita and Dexpanthenol, associated with Imipramine Hydrochloride were satisfactory. Two years later, there was no relapse of the clinical picture in both patients, even without any treatment.

Keywords: Cheilitis; Disease, Skin; Lip

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What's already known about this topic?

Although there is no consensus about its pathophysiology, clinical course and treatment, Exfoliative Cheilitis is a rare and an inflammatory skin disease which affects the lips. And it can cause functional and aesthetic problems, being associated with psychiatric and/or emotional disorders.

Whats does this study add?

The used treatments were effective, achieving the regression of the injuries, clinical improvement and weight gain, impacting on the patients' life.

INTRODUCTION

Exfoliative Cheilitis (EC) is an inflammatory disease which affects region between the skin and the mucosa of the lips¹⁻⁹, mainly the lower lips^{1,6,8}. It can be presented as erythema, irregular desquamation, lip fissures and haemorrhagic crusts^{1,2,5-9}. Patients affected by EC can develop functional (eating and speech) and aesthetical problems, leading to social isolation^{1,4-8}. The aetiology and pathogenesis of this condition are still unknown^{2-6,8,9}, although several studies sources relate EC either with the habit of licking or biting the lips^{1,2,4-9}, or with the pathogenic mechanism of psoriasis⁷.

This case report aims to describe two cases of patients with EC and provide to the literature information about the course of the affection and its empirical therapy.

Case Report 1

Female, 41 years-old, melanodermic, resident in Petrolina - PE (Brazil), presented a fissure on the right corner of the lower lip which appeared two years before the consultation. The injury, initially restricted, disseminated to the whole lower lip, with tenuous desquamation and cracks. Subsequently, the patient presented yellowish and painful crusts on the lower lip and desquamation on lower edge of the upper lip. The cycles between the fall of the crusts and the appearance of new crusts lasted about seven days. The disease arouses due to a distressing work environment where the patient was inserted. She referred to a slight weight loss at the beginning of the condition due to difficulty to eat. She also denied alcoholism, tabagism or other relevant medical condition.

On the last two years, the patient was submitted to several ineffective therapies (Table 1).

In the physical examination, the patient appeared to be depressive and apprehensive, presenting the lower lip covered by yellowish, thick and adherent crusts with haemorrhagic spots. The upper lip had a tenuous desquamation and erythema (Fig. 1A).

The patient was treated with topical dexpanthenol (ointment), fluid extract of *Chamomila recutita* and prednisone (40mg /day), associated with imipramine hydrochloride (75mg /day).

The treatment with prednisone presented excellent results one month after its beginning (Fig. 1B). Seven days after the suspension of the corticosteroid, with the maintenance of the topical medication, there

Table 1. Previous treatment of the cases 1 and 2.

Case 1		Case 2	
Treatment's Category	Previous used medications	Treatment's Category	Previous used medications
Corticosteroid	Prednisone (20mg) Hydrocortisone Desonide	Corticosteroid	Prednisone (20mg) Betamethasone Clobetazol (0,025%) Acetonide
Vitamins Supplement	Dexpanthenol - topic Beta Carotene Ascorbic Acid Alfa-tocopheryl Acetate Copper Selenium Zinc	Antibiotics/ Antifungals	Gentamicin Neomycin Gramicidin Ciclopirox Cephalexin Nystatin
Analgesics/Anti-inflammatories	Flogoral ® Troloxerutina Extract of Chamomila recutita	Anti-inflammatory	Nimesulide
Antibiotic	Benzocaine Neomycin	Moisturizer Antidepressants	Urea (10%) Imipramine Hydrochloride (25mg)
Oral Antidepressants	Fluoxetine (20mg) Imipramine Hydrochloride	Antiviral	Fanciclovir Aciclovir



Figure 1. A. Yellowish, thick and adherent crusts with haemorrhagic spots. The upper lip had a tenuous desquamation and erythema. B. Prednisone presented excellent results one month after its beginning C. Was recurrence of the desquamation interposed with healthy tissue on the upper and lower lips, without bleeding. D. Hyperkeratosis parakeratotic with mixed inflammatory infiltrate.

was recurrence of the desquamation interposed with healthy tissue on the upper and lower lips, without bleeding (Fig. 1C).

The results of complementary exams and biopsy are presented in Table 2 and Figure 1D, respectively. These results suggest the diagnosis of exfoliative cheilitis.

Case Report 2

Female, 33 years-old, Caucasian, resident in Filadelfia – BA, complained about an injury on the whole lower lip that appeared a year ago, initially with desquamation, burning and itching, afterward evolving to yellowish crusts. The cycles lasted about eight days. The injury appeared due to a distressing situation caused by the death of a relative. She mentioned weight loss caused, primarily, by the difficulty of eating. She also reported habit of licking the lips, vespertine fever for 2 days, paraesthesia in the hands and lip bleeding. The patient denied alcoholism, tabagism and other relevant medical conditions.

In the physical examination, the patient presented sadness, in addition to yellowish and thick crusts on an erythematous background on the lower lip (Fig. 2A). After the use of prednisone for two months, the injury

Table 2. Results of the laboratory exams from the cases 1 and 2.

CASE 1		CASE 2	
COMPLEMENTARY EXAMS	Results	Complementary Exams	Results
HEMOGRAM	Normal	Hemogram	Normal
URINALYSIS	Normal	Urinalysis	Normal
CREATININE	Normal	Creatinine	Normal
UREA	Normal	Urea	Normal
GLICEMIA	Normal	Glicemia	Normal
TOTAL CHOLESTEROL	Normal	Total Cholesterol	Normal
HDL	Normal	HDL	Normal
LDL	Normal	LDL	Normal
VLDL	Normal	VLDL	Normal
TRIGLYCERIDES	Normal	Triglycerides	Normal
ASLO	Normal	ASLO	Normal
FREE TESTOSTERONE	Normal	Free Testosterone	Normal
ESTRADIOL	Normal	Estradiol	Normal
SGOT (AST)	Normal	SGOT (AST)	Normal
SGPT (ALT)	Normal	SGPT (ALT)	Normal
RAPID HIV TEST	Negative	Rapid HIV Test	Negative
RAPID SYPHILIS TEST	Negative	Rapid Syphilis Test	Negative
RAPID HEPATITIS B TEST	Negative	Rapid Hepatitis B Test	Negative
RAPID HEPATITIS C TEST	Negative	Rapid Hepatitis C Test	Negative
PATCH TEST WITH STANDARD SERIES FOR 30 SUBSTANCES	Negative	Patch Test with Standard Series for 30 Substances	Negative



Figure 2. A. Yellowish and thick crusts on an erythematous background on the lower lip. B. After the use of prednisone for two months, the injury regressed, becoming clean and erythematous. C. Was recurrence of the desquamation interposed with healthy tissue on the upper and lower lips, without bleeding. D. mucosal interruption associated with inflammation chronic lymphocytic.

regressed, becoming clean and erythematous (Fig. 2B). It was detected an inappropriate buccal sanitation.

The results of exams and biopsy are presented in the Table 2 and Figure 2C – 2D, respectively.

In the last year, the patient was submitted to several unsuccessful therapies. She is currently being treated with prednisone (40mg/ day) with progressive withdrawal, omeprazole (20mg/ day), fluid extract of *Chamomila recutita* and lip balm sunscreen, associated with imipramine hydrochloride (75mg/ day).

DISCUSSION

Although the literature suggests a higher involvement of females, recent reports evince a balanced predominance between male e females¹⁻⁹. Recent reports also agree with the literature review of Almazrooa et al. (2013)⁵, which indicates an average of 59 years-old amongst 15 patients (2/3 of females).

About the cycle of the disease, Mani and Shareef (2007)¹ described a pattern of tissue renewal lasting 10 days. In the case 1, the cycle lasted seven days and in the case 2, eight days.

Despite the unknown a etiology and pathogenesis, several studies and the case 2 indicated a relation between the disease and the habit of licking the lips^{4,5,7-9}. However, reports of Mani e Shareef (2007)¹, Roveroni-Favaretto et al. (2009)², Pigatto et al. (2011)³, Nayaf (2014)⁶ and the case 1 did not confirm that association.

The condition has a disfiguring aspect, harming the aesthetic and the functional activities, implicating social exclusion or psychiatric disorders^{1,4,5,8} and then requiring the use of antidepressants, a fact evidenced in our reports.

Several therapies are being used to treat EC, such as systemic and topical corticosteroids, antifungal, antibiotics, immunosuppressive drugs, immunomodulatory, keratolytic drugs, sunscreen, emollients, cryotherapy, laser and UVB therapies, phytotherapie, antidepressants and/or psychotherapy^{1,2,4-9}. In both cases, the used treatments were effective, achieving the regression of the injuries, clinical improvement and weight gain. Although the relapse of the injuries in the case 1 after the suspension of the prednisone, both patients benefited from the use of imipramine hydrochloride, presenting emotional improvement.

*** Following a 2-year follow-up in both patients, it was observed an improvement in the emotional profile of the patients and resolution of the condition,

The knowledge about the physiopathology and the treatment is still insufficient due to the scarce information in the literature (Frame 1). The lack of consensus, in addition to the uncountable disturbances caused by this disease, makes this report an important source of additional information about that condition and its repercussions on the patients' life.

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