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Exfoliative Cheilitis in Childhood: A Successful Treatment with Tacrolimus

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Abstract

Exfoliative cheilitis is a hardly diagnosed disease by pediatricians. We have presented this report to draw the attention of clinicians because the child is the youngest patient with complete relief from using tacrolimus ointment in the literature. We present the three-year-old child with scaled, crusted, and sensitive upper and lower lips. He was admitted to different centers from the beginning of the complaints without any relief despite different treatments. The clinicians may encounter exfoliative cheilitis in different age groups. There are some approaches to management and therapy of the disease. But consensus has not yet occurred on definitive treatment, especially in childhood.

Keywords: Exfoliative cheilitis, children, tacrolimus

Introduction

Cheilitis is an acute or chronic inflammation that affects the vermillion of the lips. It is also a cosmetic problem. There are different types of this process: plasma cell cheilitis, cheilitis glandularis, actinic cheilitis, contact cheilitis, angular cheilitis, cheilitis granulomatosa, exfoliative cheilitis, and factitious cheilitis.[1]

Exfoliative cheilitis is an uncommon chronic inflammatory type of lip disease. The etiology of the disorder is still unknown. It presents with desquamation of a thick keratin scale, sensitivity, burning sensation, and sometimes fissuring of the lips.[2]

In the literature, the reported cases are in adolescent or adult age groups.[3] Because of the rare condition of the disease, the clinicians may be overlooked. Here we report the youngest patient with exfoliative cheilitis to our knowledge in the literature. The objective of the present paper is to pay attention to the disease in the childhood age group.

Case Report

The three-year-old child presented to our child health department with the chief complaints of scaled, crusted, and sensitive upper and lower lips. The lesion first appeared six months ago and gradually increased. Burning and itchy sensation, and fissuring of the lips were added to the complaint but the pain was absent. General physical examination was normal except for a painless, crusted area

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on the upper and lower lips (**Figure 1**). The crust was loosely adherent and detached easily in most places. On the lesion, there weren't any findings suggestive infectious process. Palpable head or neck lymph nodes were absent. Also, no other oral or skin lesions were noted. Lip licking and biting associated with underlying stress and anxiety because of sibling rivalry were learned. Although daily activities like eating or speaking were sometimes difficult because of the sensitivity of the lips, did not cause serious problems such as lack of weight loss. When determining personal and family medical history any specific conditions were not found.

He admitted different centers from the beginning of the complaints. Previous treatment had included the local application of corticosteroids, antibiotics, antifungal agents, and sunscreens, also topical and systemic anti-allergic agents without complete relief. But one course of antifungal agent treatment caused only partial response.

Complete blood count, sedimentation rate, and routine serum chemistries that were investigated in previous centers were all within normal limits. Immunologic evaluation and histopathological examination weren't obtained. The exfoliative cheilitis was diagnosed with the clinical findings.

A course of topical calcineurin inhibitors (tacrolimus) ointment was prescribed with a complete response after 10 days (**Figure 2**). Also, he was advised not to do factitious behavior like biting or licking the lips. Written informed consent form was obtained from the patients.

Reported cases of exfoliative cheilitis showed that females were affected more than males.[1] But Reichart et al.[4] reported that the presence of exfoliative cheilitis was more common in male Acquired Immune Deficiency Syndrome patients. In terms of age, most of the patients were adolescents or older not in the little age group.[2] In the literature, the present patient is the youngest one to our knowledge.

The major symptoms are scaling and then being yellowish-white crust, frequently painless, which may affect just one or both lips, usually the lower. When these scales are removed, usually a normal appearing lip is revealed beneath, although there may be associated erythema and edema.[5] There are different additional complaints, such as; burning or itchy sensation, pain, ulceration, bleeding, and superficial fissure of the lips.[6] Candida infection can be added to patients with predisposition like immunocompromised situations.[7] If a secondary fungal infection is added to the lesion, a partial response can be seen by antifungal treatment like our patient.

The etiology of exfoliative cheilitis is still unknown although several factors are being considered that triggered the onset. Self-damaging behavior, sometimes done unconsciously may be seen in some of the patients.[8] These habits may be a sign of stress, anxiety, and depression situations. Depression, anxiety, and personality disorders have been reported commonly in association with factitious exfoliative cheilitis.[3,6,8] These activities also may be seen at factitious cheilitis, which is a different, self-inflicted entity. But most patients have factitious cheilitis and deny these habits. Our patient licks and bites lips starting with stressful period because of sibling rivalry. We think that the situation may be the factitious type of exfoliative cheilitis like the previous report.[2] Allergy is considered a possible cause of exfoliative cheilitis. Pigatto et al.[9] reported an exfoliative cheilitis case because of a dental implant. When viewed from this aspect, our patient has no allergic history of food or anything else.

The diagnosis of exfoliative cheilitis is based on the history and clinical findings. Laboratory tests and pathological examinations aren't obtained for the diagnostic workup. Histopathological findings are usually nonspecific.[8] If it was carried out, parakeratosis or hyperkeratosis, benign epithelial hyperplasia, acute or chronic inflammation, and fibrosis can be seen.[3] A swab culture can be taken if a suspected infection exists. Also, patch testing can be performed if allergic etiology is considered. Nevertheless, the diagnosis of exfoliative cheilitis is still based on the history and the clinical findings without any laboratory or histopathological examination.

Spontaneous improvement has been reported.[5] But it often has recurrent episodes.[2,6,8] Due to a lack of consensus on treatment, the clinicians may prefer different agents, such as; corticosteroids, keratolytic agents, sunscreen antibiotics, antifungals, tacrolimus ointment, or combination treatment.[3,5] In the case reported, topical Calendula officinal is ointment known as common marigold or pot marigold was used for

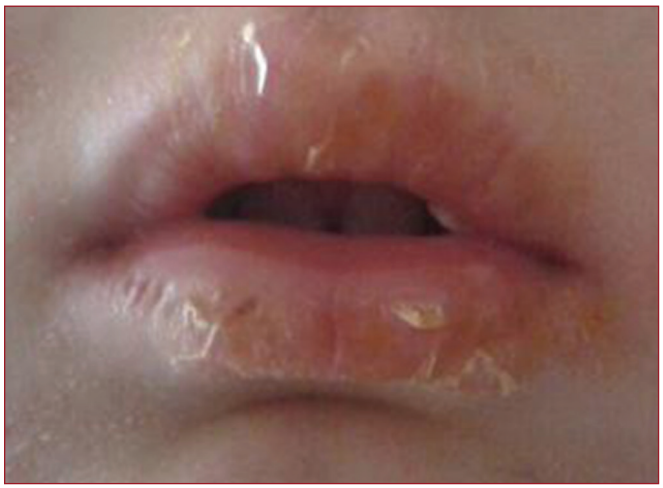

Figure 1. Scaling and crusting lips

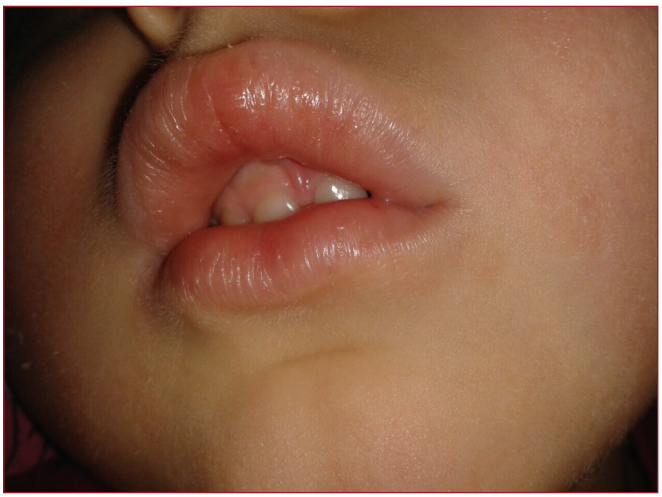

Figure 2. Treatment after tacrolimus

treatment, with complete relief.[10] Medication with antidepressants was helpful to partial response for patients with underlying depression.[2,3,6] Almazrooa et al.[3] showed all patients had complete relief and used calcineurin inhibitors, the most frequently used agent in that report. Tacrolimus is a drug isolated from streptomyces tsukubaensis, and has an effect as calcineurin inhibitors. It binds to specific receptors on T-cells to increase intracellular calcium and create an effect on several genes to change cytokines.[11] Oral tacrolimus has been used for different diseases such as preventing organ rejection in kidney and liver transplant patients.[12] As an ointment, tacrolimus is used to treatment of lupus erythematosus, vitiligo, and exfoliative dermatitis.[13-15] It suppresses inflammation like glucocorticoid agents with much fewer side effects without disturbing the collagen synthesis. A burning or itching sensation is the most common side effect, especially with application on a large surface.[16] Our patient used different agents for this complaint; corticosteroids, antibiotics, antifungal agents, sunscreens, also topical and systemic antiallergic agents. But the symptom persisted. In 10 days, a complete response was seen with topical tacrolimus treatment.

Conclusion

Exfoliative cheilitis is an unusual condition in childhood, which clinicians should be aware that is a clinical diagnosis without necessity further investigation, and tacrolimus ointment is an effective and safe drug for treatment resistance exfoliative cheilitis cases.

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Informed Consent: Written informed consent was obtained from the parents of the patient.

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