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Exfoliative Cheilitis: A Case Report

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Introduction: Exfoliative Cheilitis (EC) is a rare chronic inflammatory condition affecting the vermillion border of the lips and characterized by excessive production of keratin. In this paper, a case of chronic EC that was relatively resistant to usual treatment is to be introduced. Also, we explain its clinical course and treatment procedures and measures.
Case Presentation: We report a 25-year-old Iranian woman presented with desquamation of her lips. History, clinical feature and histopathological examination revealed a diagnosis of EC.
Conclusions: Although treatment of EC in the present study had considerable success in the short-term follow-up, it should be noted, due to the unknown of the exact cause, no specific treatment or protocol has still been identified.

Keywords: Cheilitis; Lip; Case Report

1. Introduction

Cheilitis is a term that refers to inflammation of the lips .This may comprise inflammation of the skin around the mouth, the vermilion border and/or labial mucosa, but vermilion border is more commonly involved (1, 2). Cheilitis include the following types: Chapped lips, actinic cheilitis, angular cheilitis, eczematous cheilitis, infectious cheilitis, granulomatose cheilitis, drug-related cheilitis, exfoliative cheilitis, plasma cell cheilitis and cheilitis glandularis (3).

Exfoliate Cheilitis (EC) is a rare chronic localized inflammatory condition of the vermillion border, which is characterized by the regular shedding of surface keratin layer. Vermilion is the junctional zone between the skin and mucosa, where has a thick squamous epithelium and rich capillary network (4-6). Symptoms of EC are tenderness and burning lips with different intensities. Patients may avoid participation in society due to inappropriate appearance of lips (4-8). The cause of EC isn't known, and yet there is no effective therapeutic intervention for it (8, 9). Numerous treatments with variable efficacy rates were suggested for the management of EC. Topical treatments include antibacterial and antifungal ointment, corticosteroid ointment, sunscreen, petroleum jelly, herbal product, urea 20% ointment, tacrolimus ointment, salicylic acid ointment and cryotherapy systemic treatments consist of corticosteroids, antifungal and antidepressants (5, 8-10).

2. Case Presentation

The patient was a 25-year-old woman referred to the oral medicine department, dental school, Hamadan University Medical Sciences. Her chief complaint was desquamation of her lips along with the itching and dry skin around her mouth. Lower lip was involved one year ago and upper lip was affected for one month. Clinical course of the lesion was in a manner that yellowish white crust is formed on the lips in a erythematous bed then erythematous area slowly decreased after that, within seven to ten days, the keratinized crust becomes thicker and its connection to the underlying tissue is weakened so that the patient can remove them easily and this cycle begins again. This situation has made concern and inconvenience in patient and has decreased patient's presence in the community and she had no gastrointestinal symptom or any kind of underlying disease. Recently, she had not used new toothpaste or cosmetic cream around her lips, especially coinciding with the start of the problem. This problem was not seen in any of the patient's family members. The patient had a habit of licking and biting her lip one year ago, which has left it recently. The patient was treated intermittently and discontinuously with flucinolone cream 0.025%, betamethasone cream 0.05%, metronidazole tablet 250 mg, ketoconazole tablet 200 mg, prednisolone tablet 5 mg and diphenhydramine tablet 25 mg during several visits to different

physicians within the past year. By taking some drug listed above, slight improvement has appeared but it is exacerbated by discontinuation of them (Figure 1). In general examination, patient's weight and height were 56 kg and 161 cm, respectively and she had a healthy appearance. There was no palpable lymph node in the head and neck region. The lower lip was involved with more intensity and extent. A piece of crust on the lower lip surface had loose its connection and during the examination was separated easily, the underlying mucosa was erythematous with no fissuring or papules and there were no palpable nodules in sub-mucosa. In intraoral examination, the significant plaque was on the labial surface of anterior teeth in both jaws, but the other teeth had no obvious plaque or calculus. According to the patient statement, due to brush contact with her lips and feeling pain she was unable to observe oral hygiene in the anterior part of the mouth. Other areas of the oral mucosa were quite intact and healthy. In para-clinical examination, blood test results such as complete blood count, liver function test and thyroid function test were normal. Inflammatory bowel disease was ruled out by a gastroenterologist. After removal of the superficial crust, a wedge biopsy from the lower lip was done under local anesthesia. In histopathological examination, epithelium acanthosis, hyperkeratosis and mild inflammation of the underlying connective tissue were seen. Generally, all research findings proposed a diagnosis of CE. To rule out candidiasis, the patient took oral antifungal agent (nystatin); however, there was no change in her condition. On the next visit, the patient was prescribed topical hydrocortisone 1%, Eucerin emollient cream with urea 10% and lip balm during the consultation with a dermatologist. Relative amelioration was achieved at one month follow-up. Fluoxetine 10 mg daily and alprazolam 0.5 mg daily were added to the previous drug regimen after consultation with a psychiatrist. There was a significant improvement after three months and the patient is still under follow-up (Figure 2).

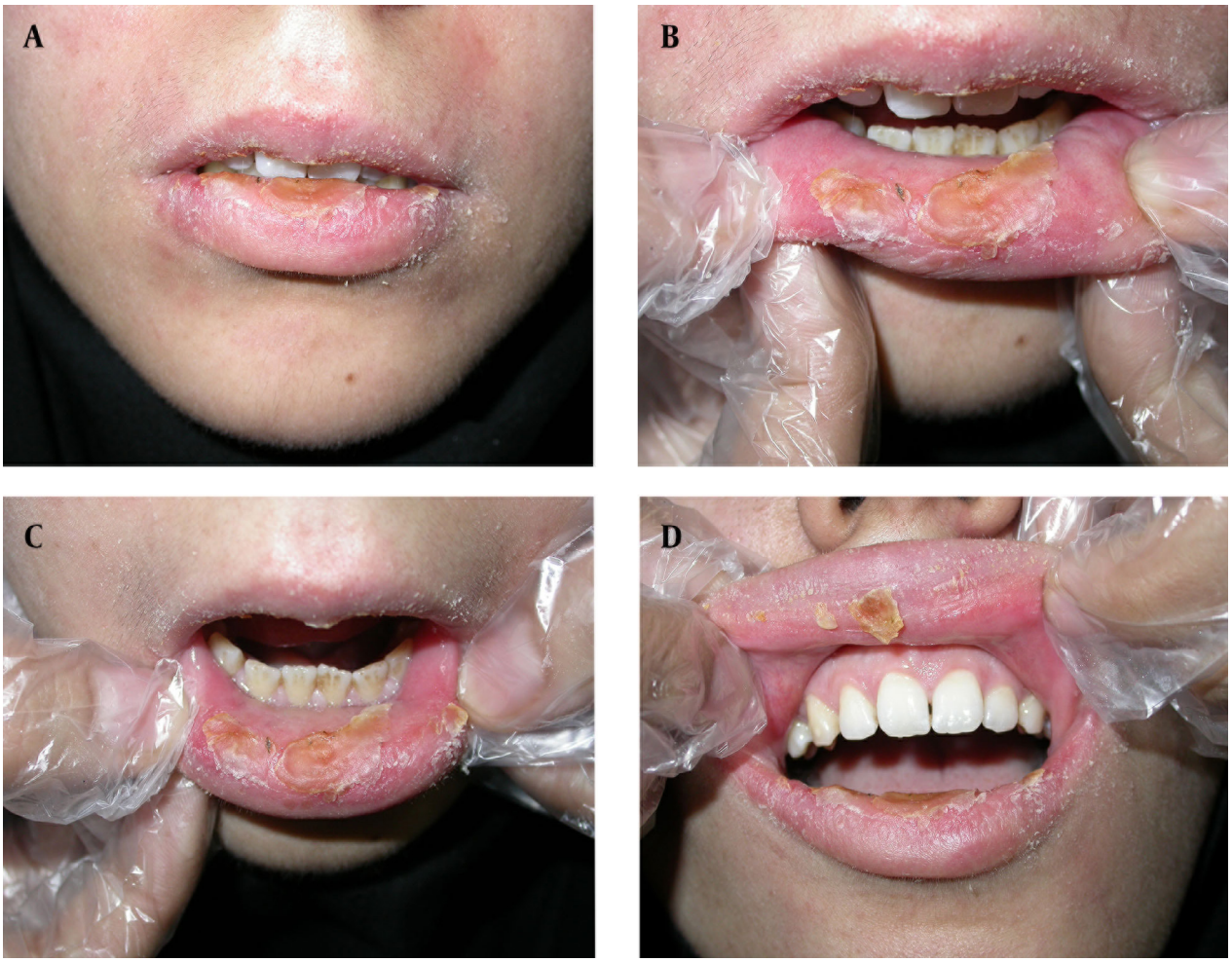

Figure 1. A 25-Year-Old Iranian Woman With Desquamation of Lips

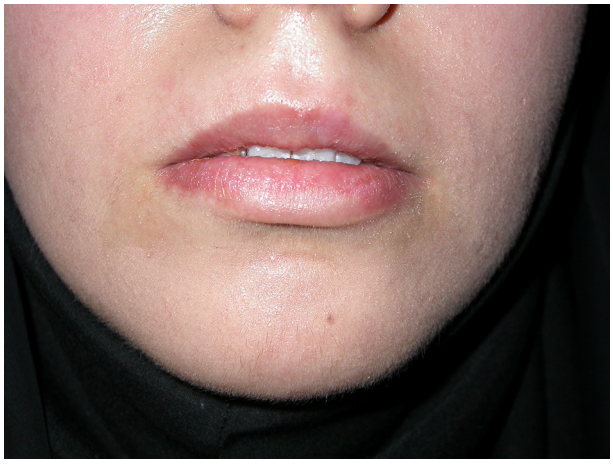

Figure 2. Significant Improvement After Three Months Follow-up

3. Discussion

Less than 200 cases of EC have been reported in the literature (51 cases in the English literature) and their diagnosis was totally based on history and clinical feature (5, 7-18). Exfoliate cheilitis is commonly seen in women and people younger than 30 years of age (4-6, 13). Both of them are consistent with our case. In many cases like our patient, lower lip is involved with more severely (6, 10-13). In a review of 48 patients with EC, 87% exhibited psychiatric condition such as anxiety and depression and starting this condition was often associated with a stressful period in a person's life (5-8, 12, 14). The history taken from the described patient and psychiatrist's consultation has confirmed the patient's depression. Also, in a review of 48 patients with EC, 47% demonstrated an abnormal thyroid function (9). Although our patient had no thyroid disease, the cause of the EC is unknown; however, many reports have described factitious activity as a cause of EC. In a retrospective study, para-functional habit of lip licking is reported in 53% of EC cases. The studied patient had a habit of licking and biting her lips one year ago, which has left it recently. She had no other risk factors such as excessive sun exposure, cold weather, mouth breathing, fungal or bacterial infection and smoking that has been noted in some studies (19). Some of the signs and symptoms related to EC include a sense of tingling and itching, pain, feeling of dryness, ulceration, fissuring and bleeding lips (7, 10-12). In the present case, only the itching sensation of the lips and dryness of perioral skin were reported and there were no other sign and symptoms. Moreover, EC may be misdiagnosed with other condition that affect the lips like atopic cheilitis and contact cheilitis, these conditions are characterized by the signs such as erythema, dryness, scaling and fissuring. These conditions are caused mainly due to cosmetic and also dental materials (2, 20). Exfoliate cheilitis may improve spontaneously. However, if it is refractory it is often resistant to treatment (5-7, 10-12). Drugs such as hydrocortisone ointment, tacrolimus ointment, petroleum jelly, tretinoin cream, urea lotion and prednisone tablet were used to manage EC. Regardless of the method, the overall response to treatment was approximately 35% in the literature. Different studies have not confirmed beneficial effect of treatment with corticosteroids, antimicrobial agents, petroleum jelly, sunscreen and dietary supplements such as iron and folic acid. The use of topical calcineurin inhibitors and moisturizing agents in the treatment of EC were associated with clinical improvement (5, 7, 8, 10, 12, 13, 18). Also, partial response to antidepressant agents such as amitriptyline, sertraline, diazepam and fluvoxamine was seen in all patients who were prescribed (5-7, 16). In this report, prescription of fluoxetine 10 mg daily and alprazolam 0.5 mg daily with hydrocortisone 1%, lip balm and Eucerin emollient cream (10% urea) together caused a significant improvement in the follow-up period of 4 months. Urea is produced naturally in the skin and causes moisture absorption and helps to rehydration of dry and scaly skin. Furthermore, urea in the Eucerin emollient cream (10% urea) penetrates to the horny layer of skin and increases the skin's capacity to absorb moisture. Eucerin covers skin surface as oil layer, which prevents water evaporation from this surface. Side effects of urea are skin irritation such as burning, itching or erythema; however, in the present case there were no side effects (21).

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Authors' Contributions

Study design, manuscript preparation, patient recalls and manuscript edition: Mohammad Vahedi, Yasamin Barakian and Parastoo Sadr.

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