

Exfoliative Cheilitis: Report of a Case

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SOMMAIRE

La chéilite exfoliative est une affection peu répandue, qui appartient à un spectre de maladies touchant la zone vermillon ou le versant cutané de la lèvre. Bien que l'étiologie de cette maladie demeure inconnue, divers facteurs – dont le stress et certains états psychiatriques – ont été associés à son apparition. Il s'agit d'une affection invalidante, qui nuit à l'esthétique ainsi qu'aux fonctions normales comme l'alimentation, l'élocution et le sourire. L'absence de traitement précis fait de la chéilite exfoliative une maladie chronique qui altère radicalement la vie d'une personne. Cette étude de cas propose un examen plus approfondi de l'évolution clinique de la maladie et présente des illustrations détaillées de sa nature cyclique.

Pour les citations, la version définitive de cet article est la version électronique : www.cda-adc.ca/jcda/vol-73/issue-7/629.html

Cheilitis is a general term that refers to an inflammation of the vermilion border of the lips.¹ The vermilion zone is the junction between the skin and the mucosa. This zone has a thick squamous epithelium and an abundant capillary supply within the interdigitating rete ridges and dermal papillae. The capillary supply makes the zone red. Cheilitis, aside from being cosmetically disfiguring, can compromise daily activities like eating and speaking.¹

Cheilitis is classified into various types: angular cheilitis, actinic cheilitis, contact cheilitis, plasma cell cheilitis, cheilitis glandularis, cheilitis granulomatosa, exfoliative cheilitis and factitious cheilitis.² Lip lesions can be manifestations of systemic diseases, a localized expression of dermatologic diseases or a localized condition of the lips.¹ In most cases, a good history, thorough clinical examination and relevant investigations will help the clinician arrive at a diagnosis.

Exfoliative cheilitis, a rare, localized condition, is a chronic superficial inflammatory condition that is characterized by regular peeling

of a superficial excessive layer of keratin.^{1,3} Bleeding may occur, resulting in hemorrhagic crusts.¹ People with this condition may have some degree of pain^{1,3} and difficulty speaking, eating or smiling.⁴ Because of their unpleasant appearance, people with exfoliative cheilitis may avoid socializing, seek periods of seclusion and be subject to clinical depression.⁴

This paper reports the case of a patient with exfoliative cheilitis and provides a photographic record of the changes that occurred over a period of 10 days.

Case Report

An 18-year-old Malay male was referred from the department of dermatology to the dental clinic of Hospital Universiti Sains Malaysia. The patient's chief complaint was pain, ulceration and crusting of the lips, which he had had for 1 year. The lesion first appeared a week before his final secondary school examinations. The patient reported that the skin over the lip thickened gradually over a 2-week period and subsequently



Figure 1: Preoperative presentation of the lips.



Figure 2: Erythematous appearance of lower lip after removal of the keratin layer (day 1).



Figure 3: A decrease in erythema is evident; the biopsy wound is seen in the centre (day 3).

became loose, causing discomfort. Once he peeled away the loosened layer, a new layer began to form again, with no complete relief from the symptoms. Subsequently, the patient refused to pursue further studies, withdrew from all social activities, and spent most of his time at home. He also indicated that he had lost weight over this period because of loss of appetite and difficulty eating.

When the patient was referred to the dental clinic, he had been taking 30 mg prednisolone regularly for 2 months, as prescribed by the dermatologist. He had also completed a course of cloxacillin and metronidazole. Results of a previous oral swab of the lesion revealed normal oral flora. He had no symptoms of gastrointestinal disturbances or other relevant medical conditions. No member of his family had a similar condition. He reported occasional lip biting, and no use of new creams, toothpaste or cosmetic items around the lips before the problem began.

The results of a general examination revealed a thin young man who weighed 42 kg. He had no fever and looked generally well. Examination of his head and neck revealed no palpable cervical lymph nodes. Most of his lower lip and some of his upper lip was crusted over, which restricted his ability to open his mouth because of pain (Fig. 1). The superficial crust on the lower lip was loosely adherent, and on removal, exposed a uniformly erythematous area with no fissuring or papules (Fig. 2). Palpation revealed no submucosal nodules. Intraoral examination showed poor oral hygiene: the patient had severe gingivitis in the anterior region, calculus deposition in the posterior teeth, multiple deep carious lesions and remnants of the roots of the lower first permanent molars. The rest of the oral mucosa was otherwise normal.

The results of a battery of tests, including complete blood work and liver function tests, a Mantoux test and chest radiograph showed no abnormalities. Results of a lip swab taken for *Candida* were negative. A wedge biopsy of the lower lip was done under local anesthesia after removal of the superficial crust. Histopathological examination

revealed parakeratosis and slight acanthosis of the epithelium, a mild-to-moderate lymphoplasmacytic infiltration of the lamina propria, and some degree of vascular dilatation and engorgement in the submucosa. Inflammatory bowel conditions were also ruled out after consultation with a gastroenterologist. The overall findings suggested a diagnosis of exfoliative cheilitis.

The development of the lesion was observed over a period of days after the removal of the superficial yellowish white crust and the subsequent biopsy of the lip. The patient was asked to admit himself to the ward so that he could be closely observed for any factitious activity and the development of the lesion. During the first 2 days, a few vesicular eruptions appeared at the junction of the vermilion border of the lip and labial mucosa lining the vestibule (Fig. 2). The erythema seemed to decrease by the third day (Fig. 3), and a thin layer of keratin formed over the lip (Fig. 4). It progressively thickened by the eighth day (Fig. 5) and was firmly adherent to the underlying mucosa. Fig. 6 shows the lesion as it appeared on the tenth day.

The patient was prescribed a topical antifungal agent (clotrimazole 2% cream), which did not improve his condition. Since a psychiatric evaluation revealed significant depression, the patient was prescribed antidepressants (fluvoxamine 50 mg once daily). These improved his condition somewhat, but did not resolve it completely. At the same time, he underwent thorough prophylaxis and instruction in oral hygiene, followed by extraction of the badly decayed teeth and restoration of the other teeth. However, subsequent dental appointments revealed that the patient was unable to maintain good oral hygiene.

Currently, the patient uses petroleum jelly for relief and continues to spend his time at home because of his condition.

Discussion

Exfoliative cheilitis reportedly occurs more commonly in females.^{1,5} However, Taniguchi and Kono,³ in a review of



Figure 4: Further reduction in erythema; formation of keratin layer is evident (day 4).



Figure 5: Marked thickening of the keratin layer (day 8).



Figure 6: The keratin layer of the upper lip seemingly thicker than that of the lower lip (day 10).

the reported cases in the literature about exfoliative cheilitis, showed that females are affected only marginally more often than males (13 versus 11). Including our case and the one reported by Leyland and Field,⁴ an equal number of males and females have been reported in the literature so far. Reichart and others,⁶ however, reported that AIDS patients with exfoliative cheilitis were predominantly male. The majority (62%) of patients affected were younger than 30 years of age, many of whom were younger than 20 years of age.³

Clinicians' knowledge of the clinical course of this disease is important for accurate diagnosis. Exfoliative cheilitis is an infrequently mentioned condition; details given about the course of this disease are inadequate. This report records in detail the clinical progress of the disease over a period of 10 days. The preoperative image (**Fig. 1**) shows a thick yellowish white coating on the lower lip and some loose adherent bilateral fragments of keratin on the upper lip. The mid portion of the upper lip seems to have a thin coating of keratin and appears almost normal. On day 1 (**Fig. 2**), the lower lip is erythematous after removal of the loosely adherent keratin coat; the upper lip was untouched. Since a biopsy of the lower lip was done after removal of the keratin layer, all subsequent images show the biopsy wound in the mid portion of the lower lip. However, the subsequent normal changes that occurred are visible on either side of the biopsy wound. The vesicular lesions seen at the junction of the vermilion border and labial mucosa may be due to the trauma induced by the removal of the plaque if the plaque was adherent in those areas when it was being removed; the presence of acanthosis in the epithelium would be a contributing factor. Over a period of the next 9 days (**Figs. 3–6**), the erythema seems to decrease gradually as the keratin layer seems to thicken. A comparison of the lower and upper lip shows that on day 1, the upper lip keratin layer had already begun to form and that toward day 10, the keratin layer of the upper lip seemed thicker than that of the lower lip. This observation suggests that the cycle proceeds differently for the upper and lower lip; certain areas of the

lip may be peeling while others may be just forming the keratin layer, giving an impression of continuously peeling lips.

Daley and Gupta⁷ and Brooke⁸ reported a similar cyclical pattern of disease activity. Brooke⁸ mentioned a 5-day period for completion of the whole cycle. Our patient claimed that the hyperkeratotic plaque developed and became loose over a period of 2 weeks and that he regularly peeled the plaque when it became loose because of the associated discomfort. The build-up then recurred over time.

Other signs and symptoms associated with this condition reported in the literature include a tingling sensation, pain, soreness of the mouth and throat, an itchy sensation, a feeling of dryness, ulceration, fissuring of the lips and bleeding.^{4,7–9} Our patient had pain and bleeding, but no other symptoms. In most reported cases,^{7–10} like our patient's, the lower lip was affected more severely than the upper lip.

The cause of exfoliative cheilitis is unknown, although many reports^{7–12} suggest factitious activity; others^{3,4,9} report exfoliative cheilitis without factitious activity. Our patient had no factitious activity, as confirmed from observation during his 10-day admission to the hospital. In fact, he took particular care to avoid pain and bleeding when moving his lips. However, the possibility of Munchausen's syndrome cannot be ruled out in those cases in which the patient does not give any indication of factitious activity when questioned or observed.¹³

The onset of the condition is often associated with a stressful period in a person's life,^{3,9} as was the case with our patient. Personality disorders associated with depression have been implicated in cases of exfoliative cheilitis, and antidepressants have been found to decrease the severity of the disease.^{3,4,9,10} However, most patients treated with antidepressants showed improvement in, but no complete remission of the disease. Our patient's condition improved when he was on antidepressants, but he, too, had no complete relief.

Raede and others¹⁴ discussed the possibility of cheilocandidosis. It involves compromised immunity or the presence of other obvious predisposing factors that cause candidal infection of the lips. The authors achieved successful resolution of such lesions with antifungal therapy. However, for people who have no specific predisposing factors, such as our patient and others,^{7,9} *Candida* could not be isolated from the lesion nor did the condition respond to antifungal therapy.

Oral sepsis has also been implicated as a cause of exfoliative cheilitis because it has resolved after implementation of good oral hygiene.⁸ Our patient had very poor oral hygiene, but even after oral rehabilitation, he had difficulty maintaining good oral hygiene. His condition may be the result of multifactorial causes such as oral sepsis, associated with and aggravated by stress that resulted in clinical depression.

Exfoliative cheilitis may resolve spontaneously,^{7,9} but if persistent, it is usually refractory to treatment and difficult to manage.^{2-4,7,8} Our patient did not respond to systemic steroids. Other unsuccessful treatment options attempted by others are cryosurgery^{3,7} and keratinolytic agents.^{7,9,10}

Conclusion

Exfoliative cheilitis predominantly affects both sexes under 30 years of age and typically follows a cyclical course characterized by normal or erythematous lips at one stage, which progressively thickens because of a steady, excessive formation of keratin until the lips flake or crust. The cycle completes with the sloughing of the keratin layer from the surface of the lips and occurs at different days on different portions of the lips. Lips with the thick keratin layer exposed to water tend to take up water and seem to have a thick yellowish white coating. The duration of the cycle may vary amongst patients.

No appropriate treatment has been identified for this condition because the cause remains unclear. Patients seem to have no choice but to wait for this condition to resolve by itself. ♦

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Acknowledgments: We would like to thank Dr. Ab. Rani Samsudin for his contribution in the preparation of this manuscript.



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The authors have no declared financial interests.

This article has been peer reviewed.