Case reports

Exfoliative cheilitis


Exfoliative cheilitis is an uncommon condition affecting the vermilion zone of the upper, lower or both lips. It is characterized by the continuous production and desquamation of unsightly, thick scales of keratin; when removed, these leave a normal appearing lip beneath. The etiology is unknown, although some cases may be factitious. Attempts at treatment by a wide variety of agents and techniques have been unsuccessful. Three patients with this disease are reported and its relationship to factitious cheilitis and candidal cheilitis is discussed.

Exfoliative cheilitis is a chronic condition which affects the vermilion zone of the upper, lower or, more commonly, both lips by the more or less continuous, excessive production and subsequent desquamation of thick keratin scales. The disorder is restricted to those cases not involving photosensitivity or allergic reactions (1). A review of the world literature by Reade & Sim (2) in 1986 disclosed only 179 cases, the great majority of which were reported in the Russian and European literature. There is a female gender predilection and the onset of most cases is before the age of 30 years (2). The characteristic presence of desquamating flakes of keratin (3) is sometimes reported to be associated with ulceration, fissuring, and bleeding (2, 4). Although some cases resolve, at least temporarily (3, 4), others persist for years (4, 5). There is no apparent association with other dermatologic or systemic diseases. This paper describes three new cases of this uncommon disease.

Case 1

A healthy 17-year-old man of Arab descent complained of a four-month history of scaling and flaking of the entire lower lip and the mid-portion of the upper lip. Desquamation was followed immediately by the formation of new scales which became thick within days. Smaller scales desquamated asynchronously from the vermilion, but occasionally a large scale involving most of the lower lip would form a slough. There was some variation in the severity of the condition from time to time. Initially, there had been a tingling sensation but pain, ulceration, fissuring and bleeding were denied. He denied excessive licking or biting of the lips, and he denied skin, conjunctival and genital lesions.

Examination revealed large, thick, tan-coloured scales covering most of the vermilion zone of the lower lip and parts of the upper lip (Fig. 1a). These could be detached easily and painlessly in most places, leaving normal appearing underlying vermilion without associated erythema, ulceration, serous crust or significant fissuring. The adjacent skin and labial mucosa were not affected.

Allergy testing with 30 common antigens was negative and the patient could not identify a specific initiating cause, although he thought it was worse if he smoked cigarettes. Microscopic examination of the scales revealed thick membranes of parakeratin (Fig. 1b) associated focally with numerous fungal spores interpreted to represent Candida, mixed bacteria, and foreign material.

The patient was advised to change his toothpaste and any other oral hygiene products to rule out a possible allergic reaction. There was no change in the disorder. Ketoconazole cream was prescribed for treatment of the fungal contaminant, but no change in the epithelial derangement was noted. Topical and systemic corticosteroids were unsuccessful, as was the application of Fucidin cream topically. Eventually, all forms of treatment were discontinued. The patient continued to remove the scales as they became loose, for cosmetic reasons.

The disease process was the same nine months after its onset. However, during the 10th month, the patient reported that he had to “peel” his lips less frequently, and by the 15th month the lips were considered normal. He has not had a relapse in three months.

Case 2

A 45-year-old Arab woman complained of a 12-year history of continuous scaling and flaking of the vermilion of both the upper and lower lips. There was no associated ulceration, bleeding or deep fissuring. Pain was absent, although the patient complained of mild soreness or an itchy feeling immediately following episodes of massive desquamation of large scales involving most of the vermilion of either or both lips. She did not wear lipstick. She was advised to change toothpaste but the condition remained unaltered. No other specific allergen could be identified. She had a maxillary denture, but the desquamative disorder started before it was made.

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She denied licking and biting of the lips prior to or since the onset of the disorder. She denied skin, conjunctival or genital lesions and was otherwise well.

Examination revealed dry, tan to yellow, thick scales some of which exhibited partial separation from the underlying tissue, involving most of the upper lip vermilion and patches of the lower lip vermilion. The skin and intraoral labial mucosa were unaffected. The scales could be easily and painlessly detached, leaving clinically normal vermilion beneath. There was no associated erythema, ulceration, deep fissuring or bleeding. Intra-oral examination revealed a Candida infection beneath the maxillary denture. Microscopic examination of the scales showed they were identical to those of Case 1, but included the presence of hyphae of Candida focally (Fig. 2). Nystatin cream was prescribed for the intraoral and the labial candidiasis. The exfoliative lip lesions did not respond to the antifungal therapy.

Other unsuccessful treatments included the use of keratinolytic agents (lactic acid 2%, salicylic acid 3%, glycolic acid 8%), topical corticosteroids, antibiotic creams, and petrolatum gels.

Six months later the disorder was unchanged, although there was a recurrence of the candidal infection.

**Case 3**

A healthy 20-year-old white man complained of a 7-month history of continuous, painless, patchy scaling and flaking of the upper and lower lip vermilion zones. There was no history of ulceration, bleeding or deep fissuring. Prior to the onset of the condition, the patient admitted to excessive licking and gentle biting of the lips, often resulting in recurrent chapping. These habits have been discontinued. He denied skin, conjunctival and genital lesions. No allergens could be identified.

Examination revealed partially desquamated tan scales involving most of the vermilion (Fig. 3a). These could be removed easily and painlessly leaving normal tissue beneath (Fig. 3b). Microscopic examination of the scales showed membranous strips of parakeratin without fungal contamination.

Unsuccessful treatments included the use of topical corticosteroids, topical antibiotics, topical antifungal agents, petrolatum gels, and sunscreens. Cryosurgery of the vermilion by liquid nitrogen spray induced swelling and massive desquamation. Upon healing, the disorder returned. All treatment was stopped and the patient was instructed to remove the scales as they became loose, for cosmetic reasons. The condition persists, although somewhat variably, ten months after onset. Candidal spores were found on microscopic examination of scales removed at nine months.

**Discussion**

Exfoliative cheilitis is a benign but often cosmetically unsightly condition. The three patients presented herein, the second patient reported by Reade & Sim (2), the patients reported by Postlewaite & Hendrickse (3), by Brook & Thomas (4) and by Tyldesley (5) all appear to show cheilitis associated predominantly with keratin scales, usually in the absence of ulceration. The etiology is unknown, although some cases, such as our third case, may be initiated but not necessarily perpetuated by lip licking or biting. Persistent crusting lip lesions whether serous scabs or keratin scales, that are associated with self-inflicted injury have been termed “factitious cheilitis”. Thomas et al (6), reported a variety of “crusting” lip lesions in six patients with psychiatric or emotional disorders, while Crotty & Dicken (7) reported 4 similar patients with “abnormal personality profiles”. Although the labial crusts found in some of these patients are dominated by keratin scales and could be diagnosed as exfoliative cheilitis, others appear to be dominated by ulcerative lesions inconsistent with this diagnosis. The confusion in terminology stems from the fact that “exfoliative cheilitis” is descriptive of a presumed etiology. Some cases of exfoliative cheilitis appear to be related to factitious injury and therefore could equally well be accurately diagnosed as factitious cheilitis. Other cases do not appear to be related to self-induced injury, and therefore a diagnosis of factitious cheilitis would be inappropriate.
Candidal infection of the vermilion, apart from angular cheilitis, usually presents as a hemorrhagic, ulcerative, or crusting lesion of the lower lip that responds to antifungal therapy (8). Keratin scaling is not a characteristic feature. A secondary candidal infection occurred in all three of our patients, which suggests that the keratin scales present a suitable environment for the spores and sometimes the hyphae of the fungus. Treatment with topical antifungal agents characteristically had no impact on the exfoliative process.

Actinic cheilitis (9) and cheilitis glandularis are not characterized by recurrent episodes of desquamating, thick, hyperkeratotic scales (10). Some cases of cheilitis granulomatosa may be associated with scaling of the vermilion but the lips also exhibit the characteristic diffuse swelling typically seen to contain granulomatous inflammation microscopically (10).

Many attempts at treatment of exfoliative cheilitis have failed. Topical and systemic corticosteroids were unsuccessful (2, 3, 5), as was intralesional injection of triamcinolone (4). Antifungal agents work against secondary fungal infection but do not prevent the formation of keratin scales (2, 3, 5). Topical and systemic antibiotics have failed to alter the disease (3, 5), as have the application of several different types of keratolytic agents (3). Petrolatum gels, sunscreens, moisturizing preparations and vitamin supplementation have been equally ineffective (2–5). Radiation therapy was unsuccessful in cases reported by TYLDSELEY (5) and by THOMAS et al. (case 4) (6). Cryotherapy was unsuccessful in our cases. THOMAS et al. (6) reported a cure (their case 1) using 1% hydrocortisone cream; however, the diagnosis of this case as exfoliative cheilitis is doubtful, as it is for their cases 2, 3 and 5, although their diagnosis of factitious cheilitis may be accurate. A similar argument applied to cases 1 and 2 reported by CROTTY & DICKEN (7). BROOKE (4) reported a cure following measures to improve oral hygiene but the follow-up period was only 4 weeks. POSTLEWAITE & HENDRICKSE’S (3) case spontaneously resolved after 6 months, without treatment, but recurred again an unspecified time later. Case 3 reported by CROTTY & DICKEN (7) also resolved, apparently with the use of amitriptyline and psychotherapy, but lesions of the lower lip had recurred by one year. The lip lesions of our case 1 resolved spontaneously in the absence of therapy and the lips have remained normal for three months.

Exfoliative cheilitis presents a significant cosmetic problem. It often affects young individuals who find themselves socially handicapped. This can lead to depression varying from mild to severe. The interpretation by some authors that the disease is a result of a psychological disorder relating to factitious habits (1, 2, 6, 7) may be true for some cases. However, the concept of a reactive psychological disorder occurring as a result of this disfiguring condition must also be considered, especially in those cases where no factitious habit can be identified by the patient or by the clinician.

References

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