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RESEARCH ARTICLE
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A RARE CASE REPORT OF FACTITIAL CHEILITIS

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Abstract
Factitial cheilitis is an uncommon disease mostly encountered in women with the history of psychosocial stress and anxiety disorders. It presents on the lips as continuous keratinaceous build-up, crusting and desquamation, consistent with exfoliative cheilitis. The affected areas can progress to superinfection with Staphylococcus aureus or Candida albicans. A 65-year-old woman patient presented to the clinic with a history of diffuse hyperkeratosis of the upper and lower lips that was initially suspected as an allergic reaction based on oral examination. On the histologic examination of biopsies and negative infectious workup led to the consideration of a factitial infection. In the end direct communication between the patient the appropriate diagnosis was discerned. This case highlights the importance of considering factitial cheilitis as the etiology of exfoliative cheilitis.

Keywords: Cheilitis, Factitial, Crusting lesion, Lips.

INTRODUCTION

The term cheilitis is the inflammation of the lip and many different factors can cause cheilitis i.e. angular, contact, exfoliative, actinic, glandular, granulomatous, plasma cell cheilitis, herpes simplex. Factitious cheilitis is a chronic condition characterized by crusting and ulceration that is probably secondary to chewing and sucking of the lips. It is difficult to diagnose rare type of cheilitis, thus proper steps of diagnostic procedure are necessary to determine the exact disease based on its characteristics.[5] Most of the cases are diagnosed by personal experiences and results without specific criteria for classification. [1-5]

CASE HISTORY

A 65 year-old female patient visited the clinic with the chief complaint of pain and peeling of her lips (figure 1) and difficulty in open the mouth. The lesion was noticed by the patient 7 years ago and gradually increased in size. Patient was nonsmoker and the physical examination showed a painful, irregular, firm, hard, crusted area with ulcerated upper and lower lip. Other oral examinations of skin and oral mucosa were normal. Oral hygiene was poor with multiple carious teeth. There were no

palpable lymph nodes. There was no history of any previous medication that could lead to any medical condition. CBC, sedimentation rate, and routine serum chemistries were all within the normal limits. An incisional biopsy of lesion showing chronic lymphocytic mucositis. A wound culture was positive for staphylococcus aureus. A diagnosis of factitious cheilitis was made and discussed with the patient along with a psychiatric consultation. Treatment was started with fluoxetine 10 mg daily and alprazolam 0.5 mg daily with hydrocortisone 1%, lip balm and Eucerin emollient cream (10% urea) and chlorhexidine mouthwash. Patient was also advised antibiotics, multivitamins along with oral prophylaxis and restoration of carious teeth. She refused psychiatric evaluation. We continued this treatment for a month and the results were not 100% positive but we could see the condition improve in a month.

FIGURES

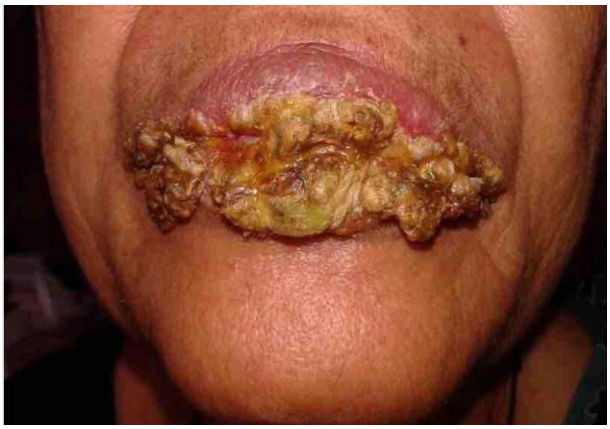

FIGURE 1: DAY 1

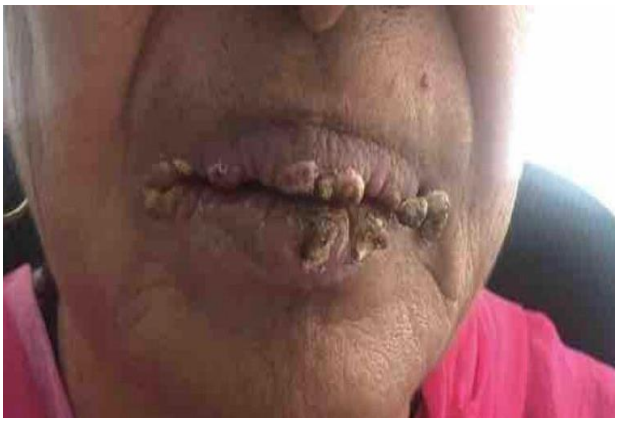

FIGURE 2: DAY 30

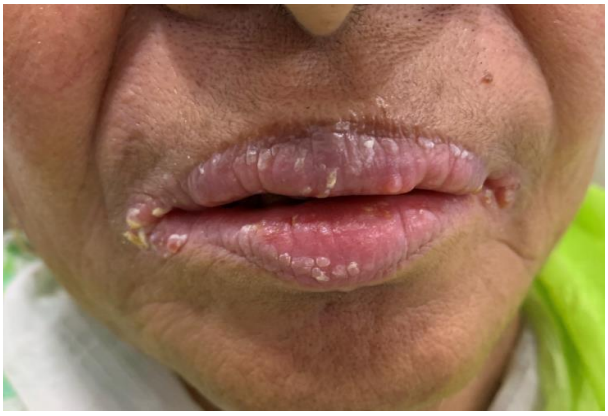

FIGURE 3: DAY 45

DISCUSSION

Factitial cheilitis is a rare condition which is characterised by cyclic and continuous peeling of excess keratin due to certain behaviours such as lip biting, persistent lip licking, sucking and picking.[6] The Self-injurious behaviour (SIB) is the damage or destruction of body tissue without suicidal intent and it's prevalence in the general population has not been established, but it is estimated that such problems could affect about 750 out of every one million individuals.[7] The exact pathogenesis of factitious cheilitis is obscure. Although factitious illness behaviour is, by definition, consciously produced, the underlying motivations for the behaviours are largely considered to be unconscious.[8] Coping deficits are widely noted for the etiology of factitious illness behaviour. Patients often have immature coping skills, not falling into any current category of personality disorder. This is consistent with observations that many factitious disorder patients come from large families or have been neglected as children, therefore lacking the nurturing conducive to the development of mature coping.[8] Patient presents with dry and scaly lips in the clinical manifestations that can have associated crusting, fissuring and hemorrhage due to the cyclical nature of the self-harming behaviours and in severe cases patient complains of pain and difficulty smiling, speaking or eating.[6] In this present case, the patient didn't approve of psychiatric intervention of any kind which is usually a typical feature of factitious disorder patients. Since she refused further psychiatric evaluation we do not have enough data to have an insight about her past, her personality, her relationship patterns and her coping skills. When asked about the family history, the patient was hesitant and failed to provide the information regarding the family members after which the patient's behaviour changed to an extent where she became anxious. Therefore, there might be an underlying family condition leading to her emotional turmoil. The role that stress and emotional problems play in the creation of lip lesions has been well demonstrated in the previous reports of factitious cresting.[9]

The diagnosis of factitial cheilitis can be even more challenging, if the patient denies factitial behaviour, as providers remain perplexed by the diffuse, cyclic, and refractory nature of the process[6] To reliably recognise and diagnose factitial cheilitis, open and honest communication between the clinician and the patient, as well as close coordination between the clinician and the pathologist is required.[6] In our case, the associated histology was rather unremarkable, with only nonspecific features identified. Conversely, the clinical presentation, as evidenced by the clinical photo, is quite remarkable and worrisome. The laboratory results and the pathology reports didn't support the organic etiology. Also, the biopsy findings weren't contributory beyond that of the physical findings.

Any patient presenting with crusting of the lips, infectious causes, contact dermatitis, and actinic cheilitis should be ruled out; then factitious crusting should be considered, and in-depth psychiatric evaluation may be necessary.[9] Also hypervitaminosis A, lupus erythematosus and licenoid dermatosis have to be ruled out. Cultures and histopathologic examination are prudent to rule out malignancy and specific infectious etiology.[8] In this case, the patient's history, the negative biopsy findings and a thorough clinical evaluation- including the psychiatry consultation – excluded the organic causes.

Depending on the presence of a comorbid Diagnostic and Statistical Manual of Mental Disorders IV Axis I disorder (e.g., depression) or a comorbid Axis II disorder (e.g., borderline personality), Psychopharmacological and psychotherapeutic treatments should be used first line according to the diagnosis. Other than targeting comorbid psychiatric disorders, there is no standard pharmacological treatment for factitious disorder. Moreover, one must keep in mind that an underlying mood or anxiety disorder which is curable, bodes for a better prognosis, whereas an underlying personality disorder, points to a poorer prognosis.[7]

In this case, the treatment was started with Fluoxetine 10 mg daily and Alprazolam 0.5 mg daily with Hydrocortisone 1%, lip balm and Eucerin emollient cream (10% urea) and Chlorhexidine mouthwash. 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.[10] Side effects of urea are skin irritation such as burning, itching or erythema; however, in the present case there were no side effects. Patient

was also advised antibiotics, multivitamins along with oral prophylaxis and restoration of carious teeth. However, patient refused the psychiatric evaluation. We continued this treatment for a month and the results were not 100% positive but we could see the condition improving in a month.
Once the diagnosis is made, management of the patient is challenging. Usually patients with this condition have underlying psychological disturbances.
Therefore, the management of factitial cheilitis is ushered on a multi-disciplinary approach that involves psychiatry, dermatology, oral medicine/pathology, and primary care physicians, for accurate, timely diagnosis and coordination of psychotherapy sessions and pharmacotherapy.[11]

CONCLUSION

Distinguishing Factitial Cheilitis from infectious cheilitis, contact dermatitis, actinic cheilitis, photosensitivity dermatoses, exfoliative cheilitis, cheilitis glandularis and neoplasia that may look similar on physical examination should thoroughly be done.[8]Comprehensive clinical history, utilization of basic laboratory tests and histopathologic evaluation are required to exclude other diseases and a thorough psychiatric evaluation and treatment is vital for successful management of these patients.[7] We strongly think that the prognosis varies based on the underlying emotional condition of the patient. Therefore, with patient motivation and thorough treatment protocol, the successful management of such patients can be positively achieved.

Declaration by Authors

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