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# Eosinophilic pustular folliculitis with labial and oral involvement: report of a rare presentation

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## Abstract

Eosinophilic pustular folliculitis (EPF) is a recurrent inflammatory dermatosis primarily involving hair follicles. Several subtypes of EPF have been described: Classic EPF, infantile EPF, and immunosuppression-associated EPF. Although classic EPF has a predilection for face, involvement of hairless areas such as palms and soles has been reported frequently. There are rare case reports of mucosal EPF. Herein, we report a woman who presented with classic EPF involving the lip and oral mucosa.

*Keywords: eosinophilic folliculitis, pustular, lip, oral cavity*

## Introduction

Eosinophilic pustular folliculitis (EPF) is an inflammatory, non-infectious disease primarily involving hair follicles. EPF usually presents as arcuate, peripherally spreading, pruritic papules and pustules. The lesions tend to recur frequently [1]. The first case of EPF was presented in 1970 in Japan [2]. Since then most cases (over one-third of cases) has been reported from Japan [3].

The histopathological examination of the lesions shows a folliculocentric, predominantly eosinophilic infiltrate in the dermis [4]. Besides the classic type of EPF there are several clinical subtypes of EPF that include both infantile EPF and immunosuppression-associated EPF (HIV-associated and malignancy-associated), [1].

EPF tends to involve hair-bearing areas of the body and is mostly seen on the head, especially in classic EPF. Nevertheless, there are several cases of EPF involving palms and soles (acrosyringial involvement), [5-7].

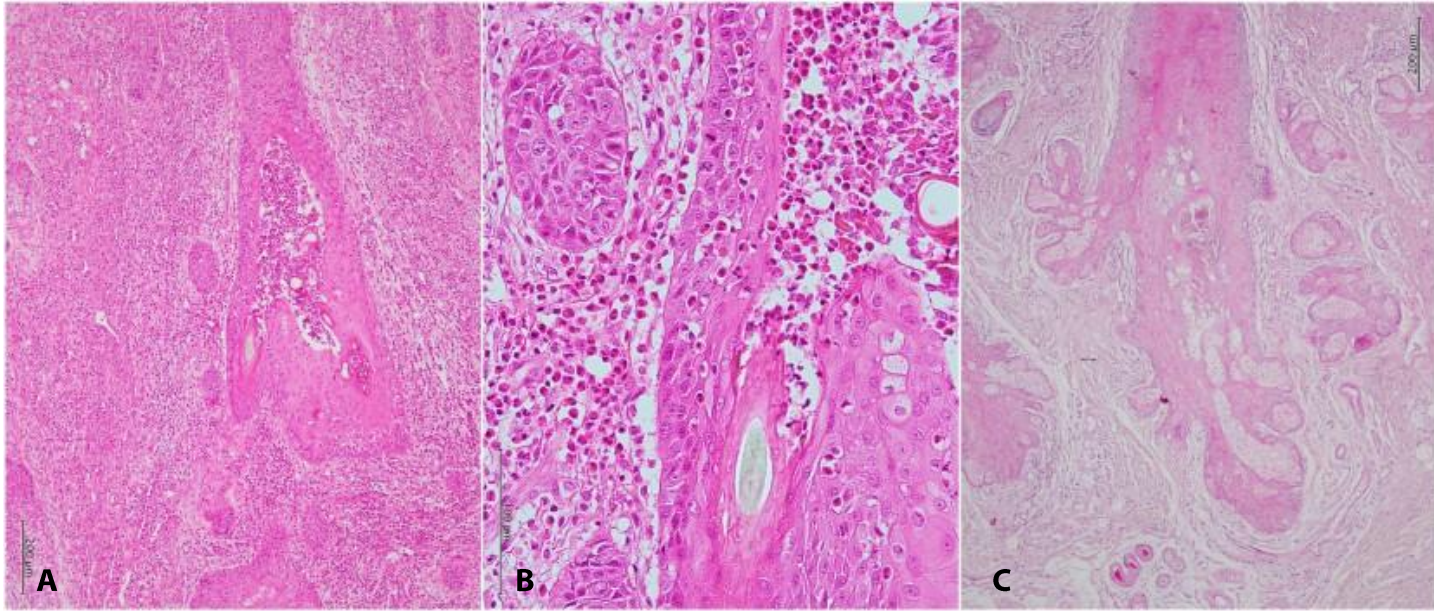
To the best of our knowledge, involvement of the oral/labial mucosa has been reported in two cases [8, 9]. Herein, we report the third case of EPF involving labial and oral mucosa.

## Case Synopsis

A 33-year-old woman presented to our outpatient dermatology clinic with a pruritic, oozing, and vegetative plaque on the chin extending to the lower lip for one week. The patient mentioned similar lesions in the past that started as multiple, pruritic,



**Figure 1.** Clinical photos. **A)** Pruritic, oozing, indurated, thickly crusted, and vegetative plaque on the chin and lower lip before treatment. **B)** Improvement of lesions after 6 days treatment with indomethacin. **C)** Relatively complete resolution of lesions after one-month treatment with indomethacin.



**Figure 2.** Histopathology. **A)** Dense folliculocentric eosinophil-rich infiltrate and follicular epithelium infiltrated by large numbers of eosinophils. H&E, bar equals 200 $\mu$ m. **B)** Perifollicular eosinophils and follicular exocytosis of eosinophils. H&E, bar equals 100 $\mu$ m. **C)** No fungal element, periodic acid-Schiff, bar equals 200 $\mu$ m.

papules and pustules on her chin and lower lip one month before the recent presentation.

No significant findings were noted in the past medical history except for chronic constipation, hemorrhoids, and gallstones leading to cholecystectomy. Drug history was not contributory.

On examination, she was afebrile (temperature 36.8°C) with normal blood pressure (110/80 mmHg) and heart rate (84 beats per minute). She appeared ill but not in distress. General physical examination showed no significant findings. There was no regional lymphadenopathy.

Dermatological examination revealed a well-defined, pruritic, oozing, indurated, thickly crusted, and vegetative plaque on the chin and lower lip (**Figure 1A**). Also, small erythematous erosive plaques were detected on the ventral surface of the tongue and floor of the mouth.

Complete blood count showed leukocytosis (WBC 12,240/ $\text{m}^3$ ) and eosinophilia (16% eosinophils). Mycological examination was negative.

The patient was treated empirically with intravenous ciprofloxacin and clindamycin for suspicion of secondary bacterial infection before receiving the negative result of bacterial culture. There was no

clinical improvement with antibiotic therapy. Skin biopsy was performed.

Considering the endemicity and clinical diversity of leishmaniasis in our region, leishmanial smear and polymerase chain reaction (PCR) was performed and the result was negative. Herpes simplex virus PCR was also negative. HIV antibody serology (ELISA) was negative.

Histopathological examination of the skin biopsy specimen revealed follicular pustules, follicular infundibulum spongiosis, dense folliculocentric eosinophil-rich infiltrate, and follicular epithelium infiltrated by large numbers of eosinophils admixed with neutrophils and mononuclear cells (**Figure 2**).

After receiving the histopathologic report, the patient was diagnosed with EPF without an obvious cause according to investigations (not definitely Ofuji type) and treatment with 50mg daily oral indomethacin was initiated. The lesions improved after one week and disappeared almost completely after one month of treatment (**Figure 1B, C**).

## Case Discussion

Eosinophilic pustular folliculitis is a recurrent, pruritic, inflammatory disorder that mainly affects

hair follicles. Although more than one-third of cases have been reported in Japan, EPF has presented in other countries as well [3]. The classic EPF is characterized by recurrent crops of pruritic pustules in an annular configuration, mostly on the face, neck, trunk, and arms. Infantile EPF typically presents as pruritic papulopustules mainly on the scalp, mostly during the first year of life. Immunosuppression-associated EPF presents as intensely pruritic follicular papulopustules either in HIV or malignancy settings [1].

An important step in diagnosing EPF is excluding other entities in the differential diagnosis such as tinea faciei, *Herpes simplex*, acne, rosacea, acne agminate, eczematous dermatitis, granuloma faciale, autoimmune annular erythema, *Demodex* folliculitis, scabies, subcorneal pustular dermatosis, and palmoplantar pustulosis. Clinical and histological differentiation from mycosis fungoides and follicular mucinosis is important [1].

Although hair-bearing areas of the skin are the major sites involved in most types of EPF, in 18% of classic EPF patients, palms and soles are involved despite

the absence of hair follicles in these sites [3]. In one of these cases, acrosyringial involvement was detected [5].

Labial and oral mucosa involvement are rarely reported. To the best of our knowledge, only two cases of EPF involving labial or oral mucosa has been reported in the medical literature [8, 9]. The first case was a German patient with involvement of oral mucosa [8]. The other one was an 11-year-old Korean girl with involvement of face and external lips [9]. Therapeutic options for EPF depend on the subtype. Indomethacin has been proposed as the first line drug for treating classic EPF [10]. Our patient improved by oral indomethacin after one month of treatment.

## Conclusion

According to this case report, EPF should be considered among the conditions in the differential diagnosis of dermatoses of hairless areas including lip and oral mucosa. This presentation further challenges the follicular-based nature of EPF.

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