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Eosinophilic Pustular Folliculitis Involving Labial Mucosa, Which Improved with Naproxen

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Dear Editor:

Eosinophilic pustular folliculitis (EPF) is a sterile eosinophilic infiltration of hair follicles. There are three variants, including the classic, immunosuppression-associated, and infantile type¹. Although they are histologically indistinguishable from each other, some propose that the three types be regarded as different disease entities due to their different responses to treatment.

Histologically, the most diagnostic feature of EPF is the infiltration of eosinophils in hair follicles and perifollicular areas¹. However, the term 'folliculitis' is being challenged², on account of the fact that approximately 20% of patients with classic EPF have the disease on palms and soles¹.

Here, we report a case of EPF involving the labial mucosa, which improved with naproxen. An 11 year-old girl presented with itching erythematous plaques and clustering pustules on the lateral side of the nose and perioral area, with erosive lesions on the external lips (Fig. 1A). A biopsy was taken from the lateral side of the nose at a district hospital. The lesion at nose showed the same feature and connected with labial lesion. It demonstrated follicular and perifollicular infiltration by eosinophils and

other inflammatory cells (Fig. 2). The diagnosis of classic EPF was made at the hospital, and she was treated with systemic prednisolone, cyclosporine, dapsone, and/or topical corticosteroid, pimecrolimus. However, her symptoms waxed and waned over 6 months and showed improvement only with the systemic prednisolone. Routine blood test was within normal limits without eosinophilia. Because pruritus was the prominent symptom rather than pain and tenderness, infectious condition was less suspected. Suspecting EPF, we started her on naproxen, 250 mg twice a day; and after three months, her symptoms greatly improved (Fig. 1B).

The etiology and pathogenesis of EPF have still yet to be fully elucidated, and there are multiple treatment options. The utilization and efficacy of therapies seem to depend on the type of EPF². Topical corticosteroids tend to be the first choice in all EPF variants¹. In children, topical calcineurin inhibitors and oral antihistamines are also usually effective and are viewed as the first-line agents³. However, our case was recalcitrant to various treatments, including topical corticosteroids and topical calcineurin inhibitor. Only the naproxen had a remarkable effect.

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Fig. 1. (A) Erythematous plaques and clustering pustules on both lateral side of the nose with central clearing and peripheral extension, involving lips. (B) Greatly improved state after treatment with naproxen.

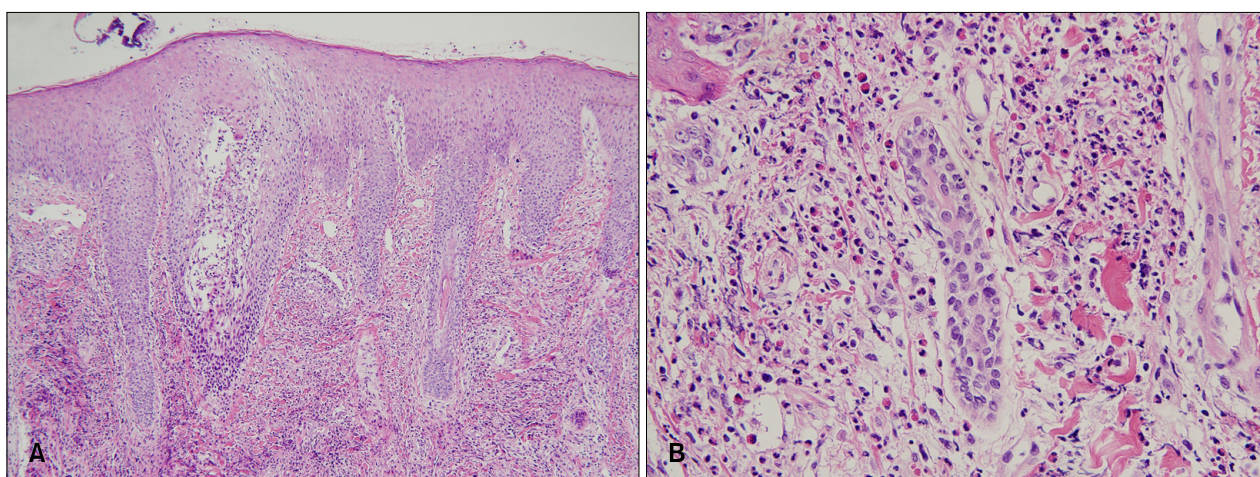


Fig. 2. (A) Inflammatory cell infiltration into hair follicle and around perifollicular area (H&E, $\times 100$), (B) perifollicular eosinophilic and neutrophilic infiltration (H&E, $\times 400$).

Nonsteroidal anti-inflammatory drugs (NSAID), particularly indomethacin, have been suggested as the treatment of choice in classic EPF². There are few reports dealing with naproxen, a type of NSAID widely being used, on EPF^{4,5}. We have reported the effects of naproxen as a first-line option on classic EPF in a Korean literature⁴. It showed 69% (11/16) of complete response rate in adult cases. This report implies that naproxen may also be safely used in children.

There has been only one case in a German literature, which involved oral mucosa in classic EPF. Our case had the labial lesions mainly on the external surface, which is

a keratinized stratified squamous epithelium rather than a true mucosa. But as there are no follicles in the external lip, as in the palms and soles, it is more likely that this disease may not be a real follicular disorder at all.

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