

Reactive Benign Follicular Mucinosis: A Report of 2 Cases

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We report 2 cases of adolescents who developed follicular mucinosis following cutaneous infections. A 17-year-old adolescent boy was evaluated for a 2-week history of erythematous papules and plaques on his face and neck. One month prior to presentation a culture was taken that was positive for methicillin-sensitive Staphylococcus aureus-associated impetigo. Biopsies from 2 representative lesions demonstrated follicular mucinosis without evidence of folliculotropism or T cell gene rearrangements. A separate case involved a 17-year-old adolescent girl who presented with an edematous plaque on her right preauricular region and scattered erythematous papules and small annular plaques over her face 2 weeks following a herpes simplex virus type 2 (HHV-2) infection on her face. Biopsy showed follicular mucinosis without evidence of epidermotropism or lymphocyte atypia. There was no herpesvirus cytopathic effect. The first case rapidly responded to an oral prednisone taper and the second case resolved over several weeks without further treatment.

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Case Reports

Patient 1—A 17-year-old adolescent boy presented with a 2-week history of erythematous papules and plaques on his face and neck. One month prior to presentation a culture was taken that was positive for methicillin-sensitive *Staphylococcus aureus*-associated impetigo located on both post-auricular regions bilaterally. The rash cleared with a short course of amoxicillin and mupirocin

ointment. Prior to presentation to our clinic he began to develop deep, acneiform, soft papules and annular plaques on his cheek (Figure 1) and neck. The lesions were not tender or pruritic and were not exacerbated by sunlight.

Punch biopsies were performed on 2 representative lesions. A superficial and deep mixed inflammatory infiltrate with abundant eosinophils was present. There was prominent follicular mucinosis with dermal mucin deposition (Figure 2). There was no evidence of notable cytologic atypical lymphocytes, epidermotropism, or alignment of lymphocytes along the dermoepidermal junction. Immunohistochemical staining with lymphoid markers showed an infiltrate consisting mostly of T cells, with strong expression of CD3 and no remarkable loss of CD5 or CD7. The CD4 to CD8 ratio was approximately 4 to 1. There was a smaller scattered component of B lymphocytes that stained with CD20; CD30 (Ki-1) was negative. Treatment options were discussed with the patient and his mother. They preferred to treat the lesions rather than to observe them for potential resolution. His lesions cleared during a 6-week taper of oral



Figure 1. Scattered erythematous papules and annular plaques on the cheek.

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The opinions and assertions contained herein are the private views of the authors and are not to be construed as official or as reflecting the views of the US Army Medical Department or the US Army at large.

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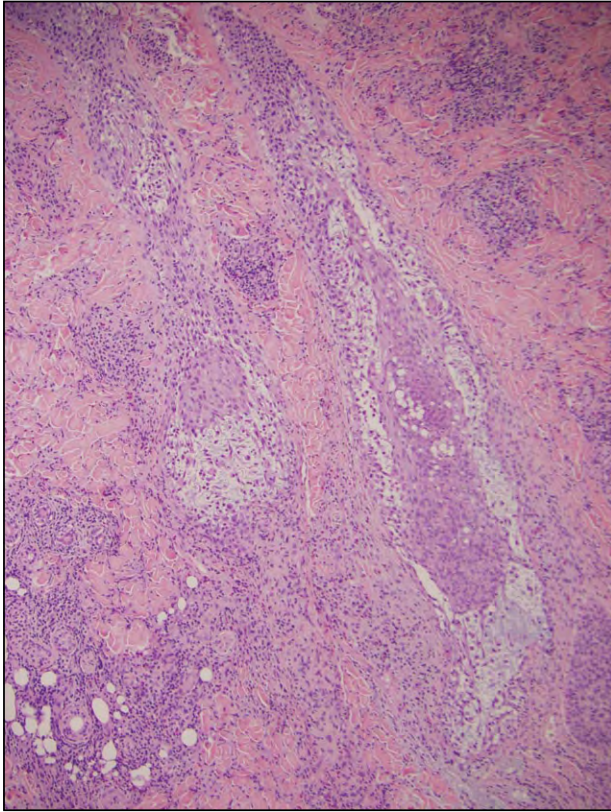


Figure 2. Punch biopsy showing prominent follicular mucinosis with dermal mucin deposition (H&E, original magnification $\times 20$).

prednisone consisting of 40 mg daily for 2 weeks, 20 mg daily for 2 weeks, and 10 mg daily for 2 weeks.

Patient 2—A 17-year-old adolescent girl was consulted for evaluation of herpes simplex virus type 2 (HHV-2) infection on her left forehead, temple, and cheek diagnosed by a direct fluorescent antibody (DFA) test. She originally presented to an acute care clinic following the development of a small collection of blisters on her left temple. She was prescribed an oral prednisone dose pack, but the lesions subsequently spread to her forehead and cheeks and eventually crusted over. Three weeks following the initial onset of symptoms she was evaluated by her primary care physician who consulted the dermatology department. The dermatologist performed a DFA test for herpes simplex virus type 1, HHV-2, and varicella-zoster virus. The DFA test was positive for HHV-2. She followed up with dermatology 1 week later for reevaluation and all of the primary lesions had resolved except for mild erythema in locations of prior inflammation.

She was seen again 1 week later with a new onset of acneiform lesions on her forehead and cheeks and a large, soft, edematous plaque on her right preauricular region (Figure 3). A punch biopsy of her right

preauricular region showed a superficial and deep perivascular infiltrate of lymphocytes and eosinophils with follicular mucinosis. No notable cytologic atypia of lymphocytes was seen. There was interface change with increased intraepithelial mucin and exocytosis of lymphocytes within several follicular infundibula. No herpesvirus cytopathic effect was seen.

Comment

Follicular mucinosis is a reaction pattern of inflammation with intrafollicular mucin deposition that often is associated with neoplastic, infectious, and other inflammatory processes.^{1,2} There are several reports in the dermatologic literature that argue the significance of follicular mucinosis and its association with mycosis fungoides (MF).²⁻⁵ Follicular mucinosis can be divided into 2 main types: follicular mucinosis associated with MF, and follicular mucinosis not associated with MF. The cases of follicular mucinosis that arise secondary to an acute infection or inflammatory process, such as the cases of our 2 patients, are unlikely to be associated with MF, have a good prognosis, and resolve over time. Cases of follicular mucinosis with clear clinical and histologic features of MF will behave similar to MF, have a poor prognosis, and do not resolve. Confusion may occur in cases that are classified as idiopathic because there is no clear trigger for the reaction pattern and a histologic confirmation of MF cannot be made. Several reports of idiopathic acneiform follicular mucinosis in adolescents have been reported.⁶⁻⁸ These cases were not associated with MF and likely represented reactive processes. On the other hand, there have been reports of idiopathic follicular mucinosis progressing to MF² with a transition time of 6 to 10 years from follow-up.^{2,3}

Differentiating between cases of follicular mucinosis associated with MF and cases of follicular



Figure 3. Large, soft, edematous plaque on the right preauricular region.

mucinosis not associated with MF can be difficult. Benign cases commonly are limited to the head and neck regions of younger patients, whereas cases of follicular mucinosis associated with malignancy can involve any region of the body and also can occur in older patients. On histologic examination, both types of follicular mucinosis showed variable lymphoid infiltrates with eosinophils. One study showed that 32% (9/28) of MF-associated cases had histologic findings consistent with a benign process.²

T-cell receptor gene rearrangements have been studied but have been ineffective in differentiating between benign and MF-associated disease. A study showed that 55% (6/11) of benign cases and 47% (9/19) of MF-associated cases had T-cell receptor rearrangements.² Other studies have shown a wide variation in T-cell receptor gene rearrangements, with no consistent pattern to differentiate benign follicular mucinosis from MF-associated follicular mucinosis.^{3,6,8}

Additionally, the presence of T cell clones were not associated with the distribution, the characteristics of lesions, or the duration of disease.³ Establishing the significance of clonality is difficult and similar dilemmas can be seen in so-called clonal dermatoses such as lymphomatoid papulosis and pityriasis lichenoides as well as inflammatory conditions such as lichen planus and lichen sclerosis.^{3,4,9}

There is no specific treatment of follicular mucinosis. A wait-and-see approach is recommended for benign reactive follicular mucinosis, as many cases spontaneously resolve within 2 to 24 months. Topical, intralesional, and systemic corticosteroids, as well as isotretinoin, psoralen plus UVA and UVA1, dapsone, antimalarial agents, indomethacin, minocycline hydrochloride, and photodynamic therapy have been reported to be beneficial.^{10,11} Cases of follicular mucinosis that are associated with MF must be treated in relation to the underlying disease.

Due to the variability of each case, we recommend long-term follow-up of all patients with follicular mucinosis to monitor for potential progression to

MF.² However, the benefit and cost-effectiveness of this approach should be questioned in cases occurring in children with clear benign sources of inflammation that precede the onset of follicular mucinosis, as it does not appear that these cases will progress to MF.

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