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Case Presentation

Diffuse lichenplanopilaris and multiple squamous neoplasms

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Abstract

Lichen planus is an inflammatory process that can affect the skin, mucosa, and hair follicles. An increased risk of squamous cell carcinoma has been noted in lichen planus of the mucosa. Rarely, in chronic, hypertrophic lichen planus of the skin, squamous cell neoplasms have been reported. We report a case of new onset lichen planopilaris with multiple squamous cell neoplasms.

Case synopsis:

A 66-year-old woman was referred for a one year history of a diffuse intensely pruritic cutaneous eruption affecting both the trunk and extremities. Prior to presenting to us, she also had developed multiple keratotic papules and plaques on the extremities that upon biopsy proved to be either squamous cell carcinomas or keratoacathomas. She had excision of five of these squamous cell carcinomas by the plastic surgery department and clear margins were obtained. However, new biopsy-proven squamous neoplasms developed in the form of papules and plaques in the vicinity of previously excised squamous cell carcinomas.

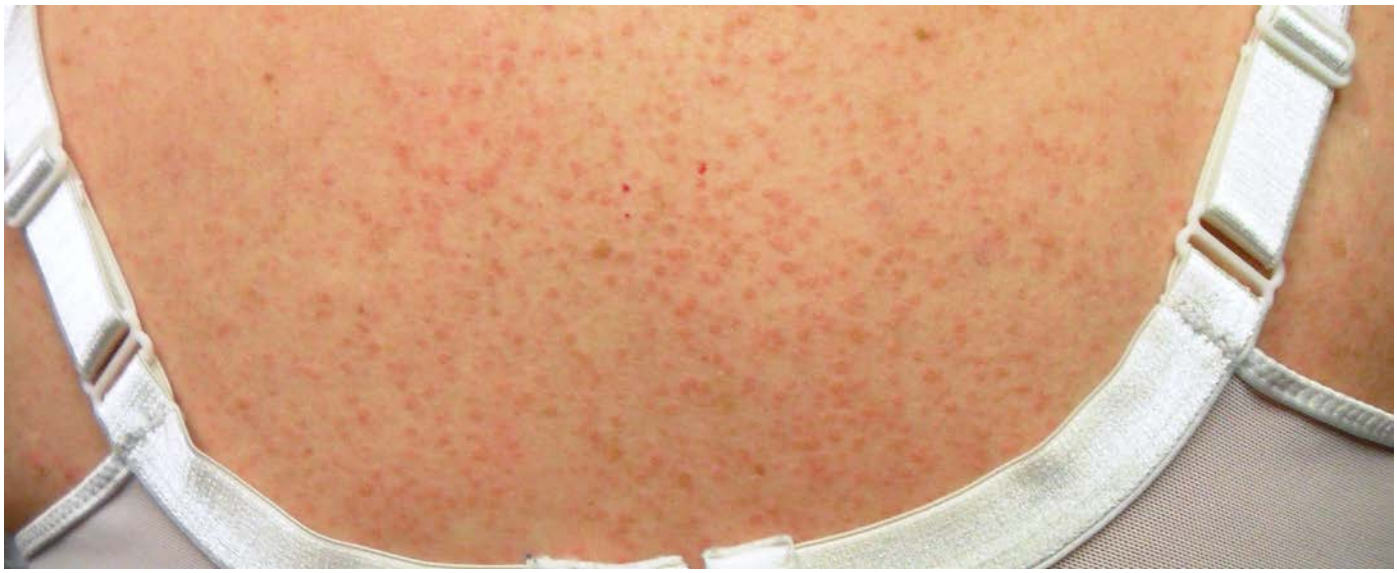

Figure 1. Numerous folliculocentric erythematous papules on the back consistent with lichen planopilaris and lichen planus.

On physical examination, the patient exhibited numerous erythematous follicular hyperkeratotic papules that were 3-4mm each on the upper arms, chest, back, and thighs (Figure 1). Multiple larger hyperkeratotic papules coalescing into plaques were present on the thighs, lower legs, forearms, and hands (Figure 2). Present on her right buccal mucosa was a lacelike whitish patch.

We performed two punch biopsies on the smaller erythematous papules. A biopsy from the left medial thigh demonstrated an interface dermatitis with involvement of the infundibula of the follicle (Figure 3). A punch biopsy from the finger identified hyperkeratosis, a lichenoid infiltrate involving the dermoepidermal junction with a few necrotic keratinocytes, epidermal hyperplasia, and a prominent granular layer. These histological findings were consistent with lichen planus.

Additional biopsies and excisions were performed on the hyperkeratotic plaques affecting the extremities. In total, there was histopathologic evaluation of fifteen biopsies and excisions of the hyperkeratotic plaques since their onset. Eleven were consistent with well-differentiated squamous cell carcinoma. A number of these squamous neoplasms were further classified as keratoacathoma (Figure 4). The other four were given the diagnosis of atypical squamous proliferation or actinic keratosis.

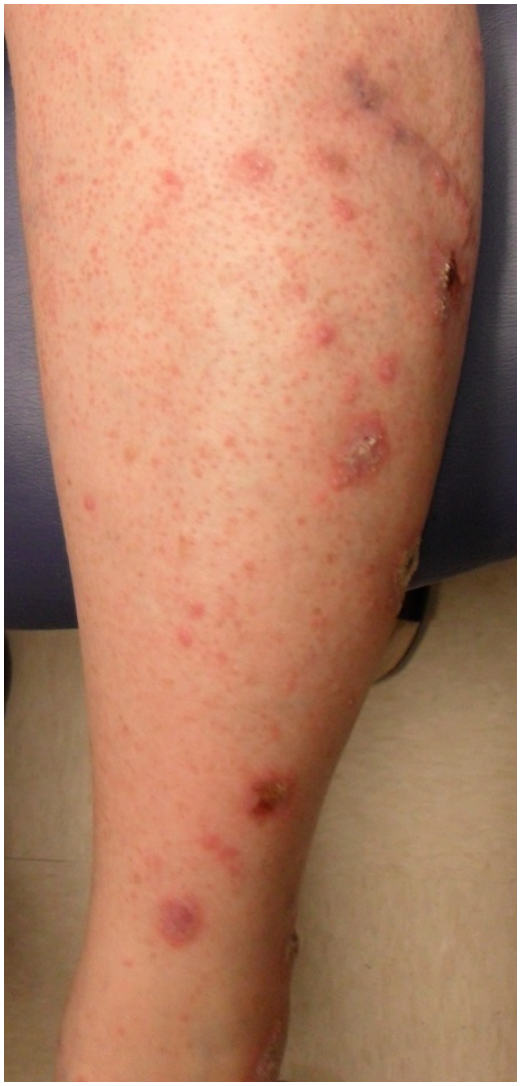

Figure 2. Multiple hyperkeratotic plaques on the lower extremity that were squamous neoplasms on biopsy.

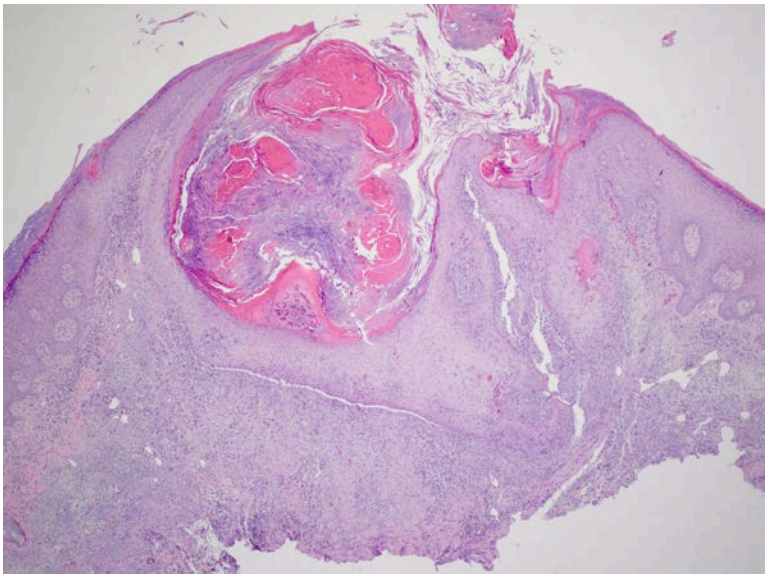

Figure 3. A biopsy from the left medial thigh demonstrating an interface dermatitis with involvement of the infundibula of the follicle and follicular plugging.

Based on clinical and pathological findings the patient's diagnoses were consistent with lichen planus, lichen planopilaris, and multiple squamous neoplasms.

She was initially started on oral prednisone and topical corticosteroids for lichen planopilaris. Acitretin 25mg three times a week was added while she was tapered off prednisone. One month after acitretin therapy was initiated, despite some improvement in the pruritic eruption, she continued to experience large painful plaques on her extremities. Acitretin 25mg was increased to five times weekly. After five months of treatment, her skin had continued to improve but inflamed erythematous scaly plaques on her legs persisted. Mycophenolate mofetil 500mg twice daily was added to her acitretin regimen to treat the inflammation. The patient decreased her dose of acitretin 25mg to three times per week owing to hair loss and mycophenolate

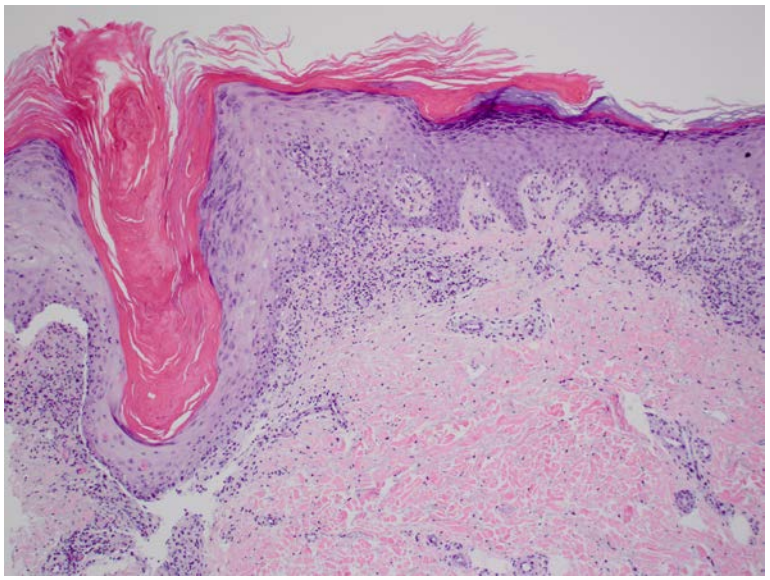

Figure 4. Well-differentiated epithelial proliferation with a keratin-filled crater consistent with squamous cell carcinoma, keratoacanthoma type.

mofetil was reduced to 500mg once daily because the patient was concerned about potential immunosuppression. Despite these dose reductions, the lichen planopilaris significantly improved on this combined regimen.

Discussion

The occurrence of squamous cell carcinoma arising in long-standing hypertrophic lichen planus has been reported [1-3]. However, the onset of multiple squamous cell neoplasms in lesions of lichen planus is extremely uncommon. In 2004, Kossard et al described two cases of hypertrophic lichen planus and one case of lichen planopilaris with co-existing squamous cell carcinomas [4](Table 1). In all three cases, the lichen planus and squamous cell carcinoma had developed nearly simultaneously and involved a localized area of skin. Histopathology showed atypical keratinocytic proliferation with prominent infundibulocystic hyperplasia. All three of these cases resolved with acitretin therapy alone. Instead of classifying these cases as variants of keratoacanthomas, the authors termed them infundibulocystic squamous cell carcinomas.

Table 1.

	Patient # 1 reported by Kossard et. al.	Patient #2 reported by Kossard et. al.	Patient #3 reported by Kossard et. al.	Patient #4 Described in this report
History	86 yo F	73 yo M	76 yo M	66 yo F
Clinical	One 5.5cm x 4.0cm plaque on left pretibial area	One 2.0cm plaque involving the columella and upper lip	Four keratotic papules that were diagnosed as SCC on bilateral lower legs. All four excised with graft repair. Keratotic nodules recurred within the bilateral grafts within one year.	Multiple follicular keratotic plaques and coalescing papules on extremities
Histopathology	SCC arising in background of infundibulocystic pseudo-epitheliomatous hyperplasia & lichenoid inflammation	SCC arising in background of lichenoid pseudo-epitheliomatous & infundibulocystic follicular hyperplasia	SCC arising in the background of lichenoid pseudo-epitheliomatous infundibulocystic hyperplasia & hypertropic LP-like reaction	Multiple SCCs biopsied & excised, punch biopsies identifying LP & LPP findings
Treatment	Resolved after 6 months of therapy with acitretin 10mg daily	Resolved after 4 months of therapy with acitretin 25mg daily	Resolved after 10 months of therapy with acitretin 25mg daily	Improved with acitretin 25mg 3x weekly & mycophenolate mofetil 500mg BID

Similar to the cases described by Kossard et al, acitretin significantly slowed the growth and resulted in regression of many of our patient's hyperkeratotic papules and plaques. Acitretin 30mg daily has been reported to be effective treatment for severe lichen planus [5]. The goal for our patient's treatment was acitretin 25mg daily. However she was unable to tolerate acitretin 25mg more than three times per week. Although our patient's condition improved, mycophenolate mofetil was added to address the intense inflammatory component of the LPP. Mycophenolate mofetil has been documented as an effective treatment option for lichen planopilaris [6].

Conclusion

Our patient and the cases described suggest that rarely there may be an association with lichen planus or lichen planopilaris onset and concurrent squamous cell carcinoma. Kossard et. al. reported patients with limited involvement of lichen planus and associated squamous cell carcinoma. The case reported above describes diffuse involvement of lichen planus, lichen planopilaris, and multiple squamous neoplasms. It seems that the onset of lichen planus and squamous cell carcinoma may be associated.

However, owing to the paucity of cases this association cannot be determined. There is speculation that the inflammation from lichen planus can cause a proliferation of the infundibulum resulting in squamous neoplasms that may histologically be defined as well differentiated squamous cell carcinomas or keratoacanthomas. Fortunately in our patient and others, these squamous neoplasms have responded to oral acitretin.

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