Superficial Mycosis Superimposing on Isolated Lichen Planus of the Lip: A Case Report and Review of the Literature

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We report a case of superficial mycosis superimposing on isolated lichen planus (LP) of the lower lip that was successfully treated. A 36-year-old woman had 2 ulcerated, scaling, erythematous plaques on the lower lip for several months with mild painful sensation. Results of histologic study showed typical features of LP, but fungal hyphae were found in the horny layer. The lesion healed with only very mild residual erythema after 6 weeks of treatment. A review of the literature shows only one noted case of tinea of the lip, which was reported in 1968 by a dental practitioner, and only several cases of isolated LP of the lip. The possible mechanism of coexistence of tinea and LP of the lip is reviewed.

Tinea (or ringworm) of the lip was first reported in the dermatologic literature in 1968 by O'Mahony.[1] It presented as a localized granulomatous lesion of the upper lip that also involved the surrounding skin, similar to tinea barbae. The fungus culture showed moderate growth of *Trichophyton verrucosum*. A short course of griseofulvin obtained a complete resolution. Since then, to our knowledge, no other tinea or superficial mycosis of the lip has been reported.

Oral lichen planus (LP) is a common disorder of unknown cause.[2] Although oral LP occurs mostly on the buccal mucosa, it also can occur on the gingivae, tongue, mouth floor, retromolar pads, and lip. Generally, LP manifests in multiple sites, and a solitary lesion on the lip is rare. We describe a case of isolated LP of the lip with superficial fungus infection.

Case Report

A 36-year-old woman with 2 persistent, painful, ulcerative lesions on the lower lip visited our outpatient department (Figure 1). The lesions had appeared 3 months prior. She had no history of skin disease or trauma to the lower lip. She also denied topical application of cosmetics, excessive sun exposure, smoking, injections, or dental work. Examination revealed 2 swollen, ulcerating, and scaling erythematous plaques symmetrically located on the outer rim of the lower lip. The overall buccal mucosa revealed no swelling, erosion, or crusting. There was no other skin lesion found inside the mouth. The upper lip was normal. On examination, no tinea lesions were noted on the nails, hair, or body. There were no signs of immune compromise, and her physical condition appeared to be very good. Laboratory studies showed normal hemoanalysis and chemistry. Tests for antinuclear antibody and venereal disease both gave negative results.

Figure 1. Two well-defined, erythematous, ulcerated, crusting plaques on the lower lip.

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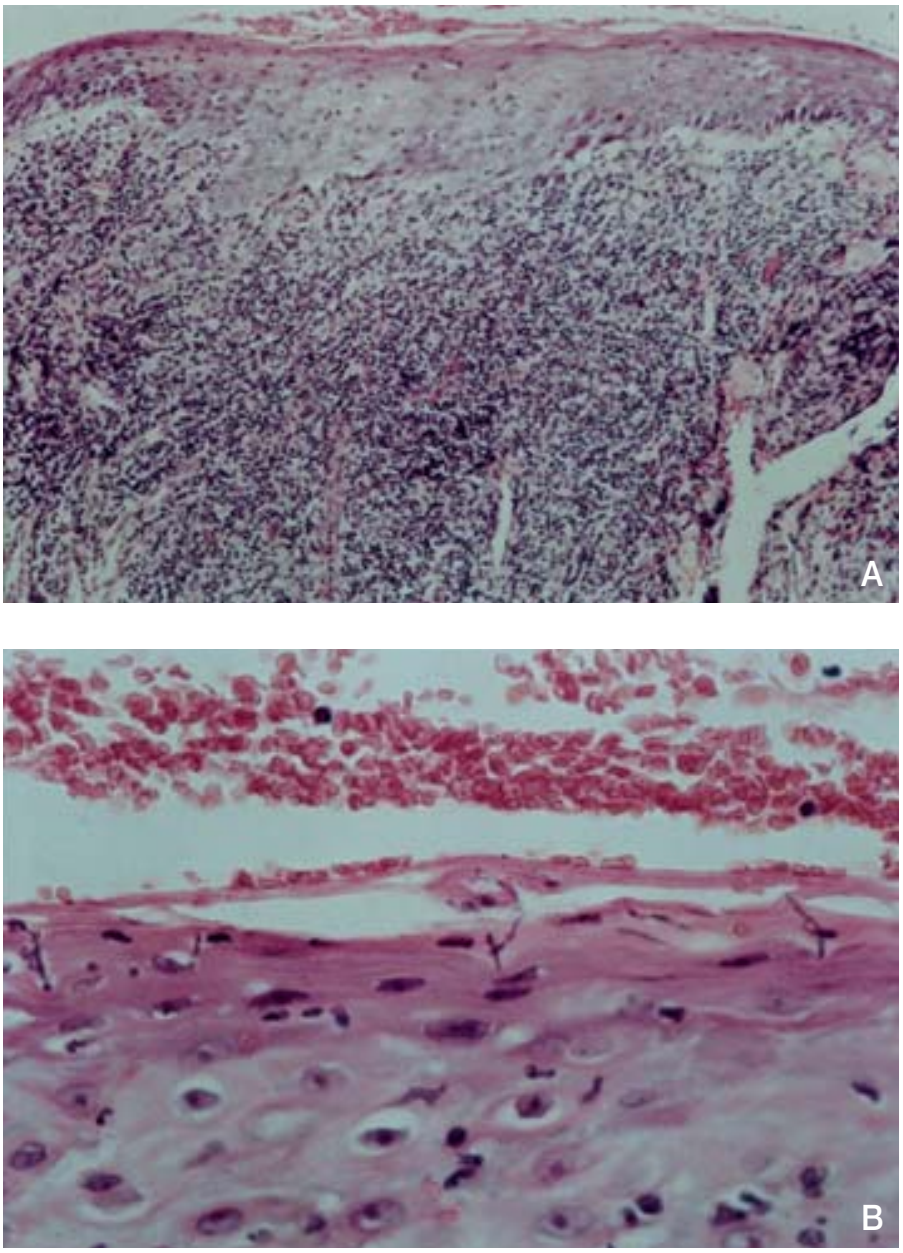

Figure 2. Typical features of lichen planus (A) and fungal hyphae in the horny layer (B).

The biopsy specimen results revealed marked hyperkeratosis, acanthosis, thickened stratum granulosum, a dense bandlike lymphohistiocytic infiltrate in the papillary dermis with basal cell liquefaction, and Civatte bodies. Several fungus pseudohyphae were found in the horny layer (Figure 2). These features are consistent with both superficial mycosis and LP.

Fungal culture of the lower lip on Sabouraud dextrose agar incubating at room temperature for 24 hours appeared as cream-colored wet colonies that were smooth surfaced. The microscopic examination revealed round-to-oval cells ranging in size from 3.7 to 5 µm by 2.7 to 4.7 µm. *Candida famata* was confirmed by API 20C AUX yeast identification system.

Treatment was started with topical 2% ketoconazole cream twice daily. However, the initial therapy course was unsatisfactory, and oral griseofulvin 500 mg/d and prednisolone 20 mg/d were given. The patient had remarkable improvement after 2 weeks. Within 3 weeks, the lesions had resolved completely. However, the lesions reoccurred one week after withdrawal of therapy. These lesions responded to topical 1% fluocinolone ointment and local intralesional triamcinolone injection.

Figure 3. Complete healing after several weeks of treatment.

No recurrence was noted after several weeks of follow-up (Figure 3).

Comment

LP is a disease of the skin and mucous membranes that frequently involves the oral mucosa. It is most prevalent in women 50 to 70 years of age. Nonspecific discomfort has been reported in most patients, and pain was noted on very few occasions. Oral LP is much more persistent, sometimes taking several years to resolve.[2,3]

Oral LP is most often distributed in the buccal mucosa. In addition, it can be found on the gingivae, tongue, floor of the mouth, retromolar pads, and lips. Multiple sites of involvement are rather common. The clinical presentations of oral LP include reticular, papular, plaque, atrophic, bullous, and erosive types. The atrophic-erosive type is most commonly encountered on the lip.[3,4]

In reviewing the literature, we only found 2 reported cases of isolated LP of the lip.[4-6] In 1961, Altman and Perry[4] reported the first case, but with no detailed description. More recently, in 1995, a well-documented case of isolated LP of the lower lip was reported by Itin et al.[5] In 1996, Allan and Buxton[6] reported a third case of isolated LP of the lip that was reticular. Ours is the third reported case involving superimposed superficial fungus infection.

The differential diagnosis includes lichenoid drug reaction, chronic graft-versus-host disease, actinic cheilitis granulomatosa, and factitious lip crusting. Oral lichenoid reaction differs from our present case for 2 reasons: (1) our patient did not take any medication prior to the onset of the disease, and (2) no amalgam were found in her teeth. Mercury sensitivity may cause lichenoid reaction.[7,8] The presentations on histologic examination of our patient were not consistent with either actinic cheilitis granulomatosa or factitious lip crusting.[9,10]

Itin et al[5] treated isolated LP with acitretin (Neotigason®), oral prednisolone, and sunscreen. Complete resolution occurred in 10 weeks. Allan and Buxton[6] applied topical steroids and established cure in 3 weeks. Our case showed resolution in 10 weeks with oral and topical ketoconazole first, then intralesional injection of steroids and topical steroids.

We propose a theory regarding this accidental coexistence of superficial mycosis and LP. The normal renewal cycle of oral mucosa is 5 days. Superficial mycosis of the lip is rare because the horny layer of the lip is too thin to provide adequate nutrients for the growth of fungi. Perhaps the ulcerated type of LP provided the host opportunity for the growth of fungus on the lip, similar to the trauma effect. In our case, the conventional treatment of LP cured the fungus infection, which also indirectly proved this evidence.

C famata, also named *Torulopsis candida*, was first isolated from air in Japan in 1922 by Saito, as reported by Meyer et al.[11] *C famata* has since been isolated from various environmental and animal sources and occasionally from clinical samples of skin or mucosa.[12-14] Human infections of *C famata* also have been described. There were 3 case reports wherein *C famata* acted as an opportunistic pathogen. In the first case, fungemia was associated with intravenous catheter insertion.[15] In the second

case, endophthalmitis occurred in a patient undergoing ocular surgery, as reported by Rao et al[16] in 1991. The third case, reported by Quindos et al[17] in 1994, occurred in a patient undergoing continuous ambulatory peritoneal dialysis in whom fatal peritonitis followed.

In conclusion, isolated LP of the lip is rarely reported. Superficial mycosis of the lip is also rarely encountered. Superficial mycosis superimposing on LP has never before been reported. We supposed that the LP lesions in the present case offered a favorable environment for the growth of fungi.

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