Oral lichen planus preceding concomitant lichen planopilaris

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Lichen planus (LP) is an immune-mediated mucocutaneous disorder with a wide array of clinical presentations. Oral lichen planus (OLP) is characterized clinically by striae, desquamation, and/or ulceration. Lichen planopilaris (LPP), a variant of LP, affects the scalp, resulting in perifollicular erythema and scarring of cutaneous surfaces accompanied by hair loss. The association between OLP and LPP has been reported previously with scant information on concomitant or sequential disease presentation. We describe a patient with concomitant OLP and LPP, and to the best of our knowledge, this is the first report on OLP preceding the onset of LPP. (Oral Surg Oral Med Oral Pathol Oral Radiol 2016;122:e82-e85)

Lichen planus (LP) is a common, immune-mediated mucocutaneous disorder affecting 0.1% to 4% of the general population.[1] The sites most commonly affected by LP are the oral mucosa and skin; however, other mucous membranes (anogenital, conjunctival, esophageal) and cutaneous appendages (nail and scalp hair) may be affected.[2] Oral lichen planus (OLP) affects females twice as frequently as males, with onset between 30 and 60 years of age and prevalence ranging from 0.5% to 3%.[3] OLP clinically presents in reticular, plaque-like, erythematous, papular, and erosive forms and may be accompanied by extraoral mucosal and/or cutaneous involvement.[3] Lichen planopilaris (LPP), also known as follicular LP, is a rare form of LP similar to OLP in gender and age distribution.[4-6] LPP affects the scalp and commonly presents as perifollicular erythema, follicular hyperkeratosis, and permanent hair loss.[4]

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The pathophysiologic mechanism for LPP is attributed to activated T-lymphocytes targeting follicular antigens, which results in failure of follicle regeneration and subsequent development of associated clinical features.[5] Various agents, such as medications, infectious agents, and contact sensitizers, have been implicated as activators of this inflammatory process.[5] Angiotensin-converting enzyme inhibitors, beta-blockers, and quinidine are medications that have been associated with LPP and OLP.[5,6] Hepatitis C virus and contact sensitizers, such as gold, mercury, and cobalt, have also been associated with these conditions.[2,3,5]

Three subtypes of LPP have been widely recognized and described: (1) classic LPP, (2) frontal fibrosing alopecia (FFA), and (3) Lasseur Graham-Little Piccardi (LGLP) syndrome.[5] Classic LPP may affect any part of the scalp, but commonly involves the vertex and typically presents as follicular violaceous erythema evolving into hair loss and subsequent scarring.[5] FFA consists of bandlike scarring alopecia of a progressive nature. This subtype of LPP predominantly affects postmenopausal women, and this may be related to hormonal imbalances, and ongoing recession of the frontal and temporal hairline is common to FFA.[5,7] LGLP syndrome is similar to OLP and classic LPP in age and gender distribution and is characterized by lichenoid follicular eruption, non-cicatricial axillary and pubic hair loss, and scarring alopecia of the scalp in a patchy distribution.[6]

Histopathologic findings associated with LPP are variable because of the progressive nature of the disorder, but is defined by destruction and fibrosis of the hair follicle.[5] A lichenoid lymphocytic infiltrate affecting the infundibulum and isthmus, while sparing the lower portion of the hair follicle, are characteristic of early lesions.[5] Degeneration of basal keratinocytes results in colloid body formation, which may also be observed on routine analysis.[6] Advanced lesions have little to no inflammation, but demonstrate extensive perifollicular lamellar fibrosis that replaces the follicular structure and bandlike scarring beneath the

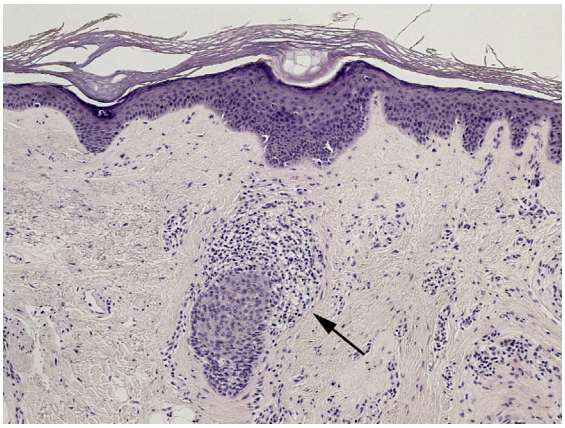

Fig. 1. A dense lymphocytic infiltrate extends along a follicle (*arrow*) consistent with lichen planopilaris (hematoxylin and eosin, original magnification ×40).

papillary dermis.[5,6] Direct immunofluorescence may also reveal positive colloid body staining with anti–immunoglobulin M (IgM), anti-IgG, anti-IgA, or C3 around the infundibulum or at the dermal–epidermal junction.[5]

The association between OLP and LPP has been reported previously, but with scant information on concomitant or sequential disease presentation. We describe a patient with concomitant OLP and LPP, and to the best of our knowledge, this is the first report of OLP preceding the onset of LPP.

CASE REPORT

A 77-year-old woman complained of generalized, asymptomatic maxillary and mandibular gingival erythema of approximately 15 years' duration (starting in 1999/2000). The patient had been previously evaluated in 2009 by a dermatologist for hair loss associated with cutaneous changes affecting her scalp. She denied any other cutaneous or mucosal surface complaints similar to her scalp issues. At that time, she had reported that her gingival condition preceded her scalp lesions by approximately 10 years. The diagnosis of LPP (most consistent with the FFA type) was established via scalp biopsy, which demonstrated a dense lymphocytic infiltrate extending along a follicle (Figure 1). Past medical history consisted of left breast adenocarcinoma (diagnosed in 2010 and treated with partial mastectomy and radiation), diverticulosis, gastroesophageal reflux disorder, and asthma. Her medications included anastrozole, albuterol, triamcinolone acetonide, and multivitamins. Of interest, the patient reported advanced progression of LPP while using anastrozole until its use was discontinued in 2015. The patient was allergic to penicillin, metronidazole, sulfa antibiotics, and latex. A systemic review was positive for joint pain in her right hand, tinnitus, and vertigo. Physical examination revealed a well-nourished, well-developed female. Examination of the scalp revealed bilateral frontal/temporal recession and scarring on the central frontal hairline with a preserved tuft of hair (Figures 2 and 3). Extraoral

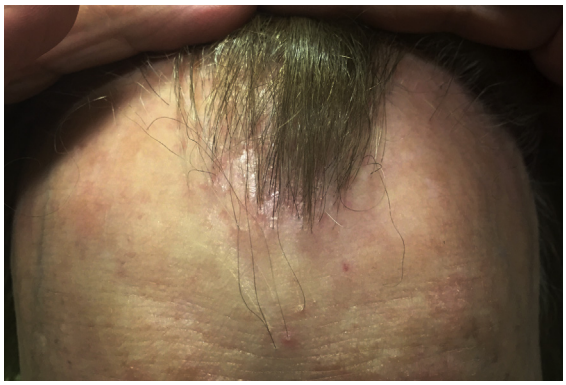

Fig. 2. Central front hair line with a preserved tuft of hair consistent with lichen planopilaris.

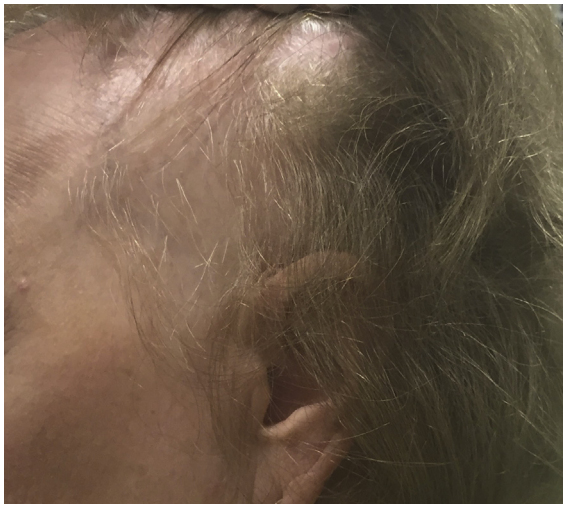

Fig. 3. Temporal hair line recession consistent with lichen planopilaris.

examination did not reveal lymphadenopathy, thyromegaly, or salivary gland enlargement. Intraoral examination demonstrated generalized erythema of the maxillary and mandibular gingivae, accompanied by striae and focal edema without ulceration (Figure 4). Incisional biopsy of the mandibular gingiva with routine tissue staining revealed parakeratotic, stratified squamous epithelium exhibiting degeneration of the basal cell layer and a dense, bandlike infiltrate of lymphocytes immediately subjacent to the epithelium consistent with OLP (Figure 5). Treatment of OLP was not recommended because of the asymptomatic nature of the condition, and the patient returned for periodic observation.

DISCUSSION

There are few reports describing OLP in association with LPP. In a retrospective study of 584 patients in the United States, Eisen identified six female patients with histologically confirmed OLP and LPP.[8] He reported the development of scalp lesions preceding the onset