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Bullous oral lichen planus: report of two cases

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Lichen planus is a common chronic mucocutaneous disease of unknown cause. Several types of lichen planus (reticular, atrophic, erosive and bullous) within the oral cavity have been described, among which the reticular and erosive types are the most common types,[1,2] while the bullous type is rarely observed.[1,3] Bullous lichen planus (BLP) lesions are commonly seen on the buccal mucosa, most frequently at the posterior areas adjacent to the second and third molar teeth. Less common localizations are gingiva and inner aspect of the lips.[4] Bullae are generally short lived and leave ulcerated lesions on rupturing.[1] The clinical diagnosis of bullous lichen planus is extremely difficult, and pathological examinations may be necessary to establish a definitive diagnosis.[5]

CASE REPORTS

Case 1

A 36-year-old female patient referred to Department of Periodontology, Faculty of Dentistry, Gazi University, Turkey, with the main complaint of painful desquamation of the gingiva for a month. The patient reported no remarkable medical history except bullous lesions that appeared apparently on the gingiva. There was no history of smoking and alcohol and drug intake. Physical examination showed no lesions on the skin. Intraoral examination revealed erythemous lesions extending from free gingiva to the attached gingiva at the premolar posterior region. The gingival tissues were generally pink and firm with slight marginal edema. Probing depths ranged from 2 to 3 mm with no detectable bleeding. There was minimal plaque accumulation, with the exception of moderate plaque. The appereances of buccal mucosa and tongue were normal. A gingival biopsy was taken for histopathological evaluation.

Case 2

A 52-year-old female patient complained of intermittently sore tongue for two months, which was related to spicy or salty foods. She had the difficulty in using her dentures since the onset of the disease. No other remarkable medical history was reported. Intraoral examination found that the lesions were located on the left side of the tongue, hard palate and anterior alveoler crests, with irregular red borders and white-yellowish pseudomembrane in the middle. The patient felt pain when the pseudomembrane was disturbed. Head and neck examination revealed no unusual findings. No evidence of conjunctivitis and skin lesions was detected. Biopsies were taken from hard palate and mucosa of alveoler crest.

Histopathologic findings

Histopathologic examinations were done at the Department of Oral Pathology, Faculty of Dentistry, University of Gazi, Turkey. Hyperkeratosis and basal cell degeneration of squamous epithelium were seen microscopically, and the separation of the epithelium from the basement membrane was noted. Compact, band-like lymphocytic infiltration was found at the epithelial connective tissue interface, and lymphocytes were present in the epithelium.

Diagnosis and therapy

Based on the clinical and histopathological findings, both cases were diagnosed as bullous lichen planus. The first patient received full-mouth supragingival tooth cleaning and oral hygiene instructions, including using a soft-bristle tooth-brush and applying the modified Bass technique. In addition, the proper use of dental floss was demonstrated. The second patient was edentulous. Both patients were treated with 40 mg prednisone each day in the early morning until the lesions were improved, and prednisone was then decreased to 20 mg and dapsone of 25 mg daily was used. Four weeks later, the oral lesions healed, and there was no evidence of new lesions.

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DISCUSSION

The incidence of oral lichen planus ranges between 0.5% – 2% among the populations;[2,6] however, the incidence of BLP is much lower.[7] BLP may be confused with other vesiculobullous diseases such as pemphigoid lesions.[8] Biopsy and histological examination are essential in obtaining a definitive diagnosis.[9] In the present cases, according to the patients' medical history and the clinical examinations, pemphigoid and other vesicullobullous lesions were suspested for differential diagnosis. Biopsies were taken and histopathologic examinations revealed the definitive diagnosis of BLP with typical features. The microscopic findings in both cases revealed separation of the epithelium from the basement membrane zone with basal cell degeneration and infiltration of lymphocytes into the subepithelial layer of connective tissue, which excluded the possibility of pemphigoid.

Most of the lichen planus lesions are reported in association with skin lesions.[10] About half of the patients with skin lesions have oral lesions, whereas about 25% presents with oral lesions alone.[11] The existence of skin lesions may be helpful to confirm the diagnosis of oral lichen planus, but not essential.[12] In the present cases, oral involvement was solely observed. Both patients had neither skin nor ocular lesions. In addition, the patients had no history of any lesions suggestive of classic lichen planus. Meanwhile, gingiva is one of the uncommon sites for BLP.[11] In the first case lesional involvement was only seen on the gingiva.

Limited information is currently available for the treatment of oral lesions associated with bullous lichen planus. Recommendations for treatment generally include the use of topical and systemic steroid medications. Dapsone has been used to treat various inflammatory and infectious dermatoses,[13] including BLP and erosive cutaneous lichen planus in both children and adults.[14,15] Both patients in this report experienced remissions, one after a course of moderate-dose systemic corticosteroids and the other after the addition of dapsone to her therapeutic regimen. Clinical improvements were seen after the treatment. The patients were followed up during treatment period, and no recurrence of the lesions was observed.

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