Oral and Vulvar Lichen Sclerosus

Lichen sclerosus (LS) is a chronic, inflammatory, mucocutaneous disorder of genital and extragenital skin (1). Simultaneous involvement of the oral mucosa is extremely rare, but it may be the only affected area (2).

A 55-year-old woman was referred to the Department of Oral Medicine, School of Dental Medicine University of Zagreb due to whitish lesions on the right ventrolateral part of the tongue and buccal mucosa with desquamative gingivitis (Figure 1, a-c). The lesions were asymptomatic but indurated on palpation. Histology was conclusive for oral lichen sclerosus (OLS). The lesions on gingiva were successfully treated with betamethasone ointment, three times a day for two weeks.

One year earlier, she had been referred to the Department of Dermatology and Venereology with progressive pruritus and dyspareunia, white patches, obliteration of the labia minora, and stenosis of the introitus (Figure 2). Histology was conclusive for vulvar LS (Figure 3, a and b). She was successfully treated for 5 months with clobetasol propionate 0.05% ointment. The patient was taking levothyroxine to treat hypothyroidism associated with Hashimoto's thyroiditis and was otherwise healthy.

Oral LS is clinically characterized by the appearance of white macules, papules, or plaques mostly appearing on labial mucosa but also on buccal, palate mucosa and on the lower lip (2,3). On the genitals, it typically manifests as atrophic white plaques, which may be accompanied by purpura or fissuring (1). While vulvar LS is often associated with pruritus, dyspareunia, and dysuria, OLS is often asymptomatic, although pain, soreness, pruritus, and tightness when opening the mouth can be present (1,2). Oral manifestations of LS, as well as association of anogenital and oral LS, are rarely reported in the literature (4-6). Tomo *et al*. searched the Medline database for papers reporting oral LS cases with histological diagnosis confirmation from 1957 to 2016 and found only 34 cases of oral LS with histopathologic confirmation of the diagnosis (4). Kakko *et al*. reported 39 histologically proven cases of OLS (2). Attilli *et al*. (5) reviewed

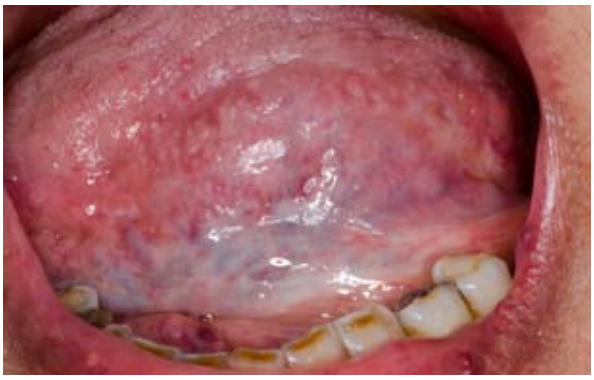

Figure 1a. Whitish lesions on the ventrolateral part of the tongue in patient with lichen sclerosus

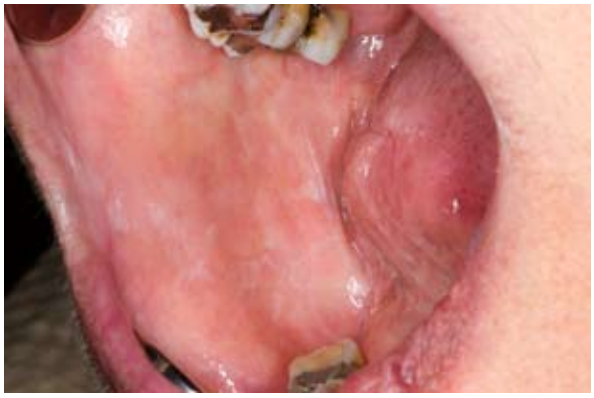

Figure 1b. Whitish lesions of the buccal mucosa in patient with lichen sclerosus

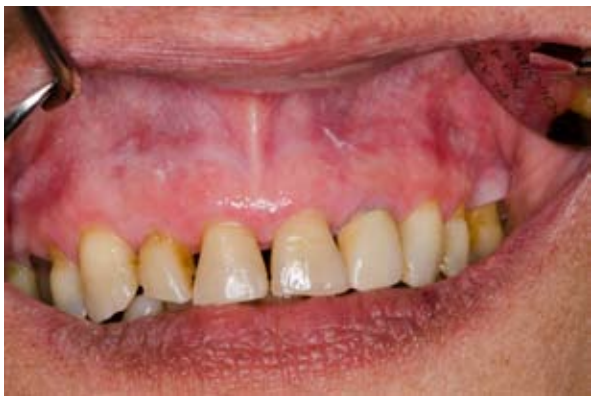

Figure 1c. Desquamative gingivitis in upper jaw in patient with lichen sclerosus

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Figure 2. White patches, obliteration of the labia minora and stenosis of the introitus in 55-year old patients with vulvar lichen sclerosus

the clinical and histologic features of 72 cases of LS with oral/genital involvement. They reported that LS was diagnosed with exclusive genital lesions in 45, exclusive lip involvement in 20, and orogenital involvement in only 7 cases (5).

Some believe that many cases of clinically diagnosed lichen planus may actually be LS and that isolated oral mucosal LS may not be as rare as is generally thought (2). While vulvar LS can occur at any age with increasing incidence with age, the median age of patients with OLS was 34 years and most of the patients were female (1,2,5).

Due to the small number of patients in the literature, treatment recommendations for OLS are not available. In case of symptomatic oral lesions, topical or intralesional corticosteroids are considered to be the first-line treatment (2). First-line treatment for anogenital LS is a potent to very potent topical corticosteroid ointment, and second-line therapies include topical calcineurin inhibitors 1% pimecrolimus and 0.1% and 0.03% tacrolimus (1). For treatment-resistant genital LS, oral retinoids, methotrexate, and possibly local steroid injections for single lesions are mainly applicable for women (1). There is limited evidence for systemic treatments for both conditions.

If it is not treated, genital LS is associated with a greater degree of scarring and an elevated risk of progression to squamous cell cancer; however, malignant transformation of OLS has not been reported (1-6).

Due to the very rare presentation in the oral cavity, it is important to notice these lesions during a dental exam.

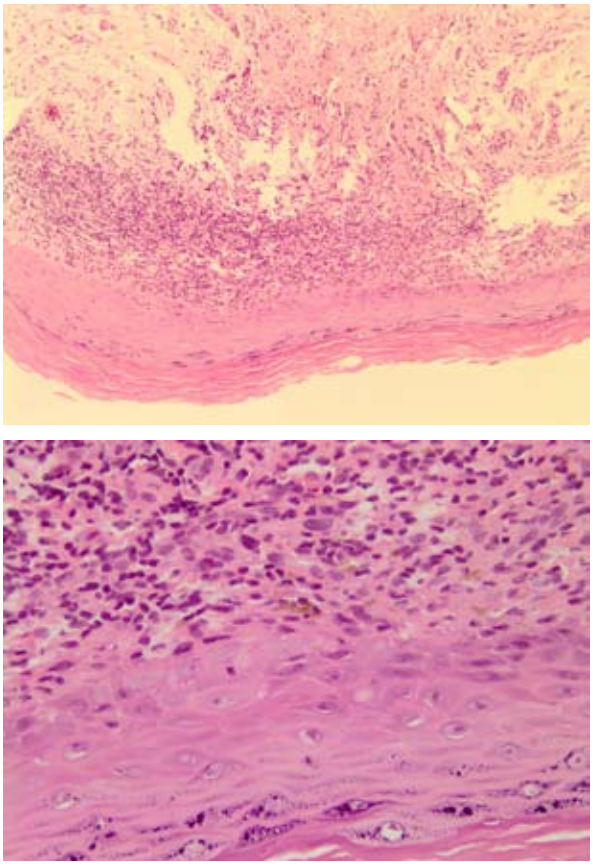

Figure 3a and 3b. Hyperkeratotic stratified squamous epithelium, partly normal thickness, partly with evidence of thinning, and with loss of the normal rete ridge. The stratified squamous epithelium shows hydropic degeneration of basal and parabasal cells in few areas. The subjacent connective tissue showed dense linear subepithelial inflammatory cell infiltrate. (Hematoxylin and eosin, 10x and 40x)

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