Lichen Sclerosus of the Upper Lip: Report of a Case, Utilizing Shikata's Modified Orcein Stain, and Review of the Literature

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ABSTRACT

Background: Lichen sclerosus is a rare chronic inflammatory disorder, affecting mainly the skin and the anogenital mucosa, while oral lesions can be the primary or the only manifestation of the disease. A rare case of oral lichen sclerosus, assessed histopathologically and histochemically, is presented, along with a thorough review of the English language literature.
Methods: A 32-year-old female presented an asymptomatic white patch affecting the skin and the mucosa of the upper lip, without other mucocutaneous involvement. A partial biopsy of the lesion was performed, along with the histochemical Shikata's modified orcein stain for elastin fibres detection. A literature review was also performed, discussing the epidemiological data, clinical presentation, and treatment modalities of all published cases with oral involvement.
Results: The histopathological evaluation revealed the presence of acellular zone underneath the basal layer of the epithelium, accompanied by deep band-like chronic inflammation. Shikata's modified orcein stain exhibited scarcity or loss of elastin fibres in the acellular subepithelial area. The diagnosis of lichen sclerosus was made. Topical application of corticosteroids was prescribed and resolution of the lesion was observed. Literature review revealed that oral lichen sclerosus is predominantly presented in females, as asymptomatic lesions of the lips and buccal mucosa; few cases have extraoral manifestations and topical corticosteroids are the main treatment intervention.
Conclusions: Despite its rarity, lichen sclerosus should be considered in the clinical and histopathological differential diagnosis of white patches of the oral mucosa. For rendering proper diagnosis, the histochemical Shikata's modified orcein stain is a useful assessment tool.

Keywords: cheilitis; elastin; lichen sclerosus; lichenoid eruptions; oral pathology.

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INTRODUCTION

Lichen sclerosus (LS) is an uncommon mucocutaneous disorder that mainly affects females over a wide age range [1]. The lesion was first described as "*lichen planus atrophicus*" in 1887 by Hallopeau; five years later, Darrier described the same lesion as "*lichen planus sclerosus*", and, in 1940, Montogmery and Hill used the term "*lichen sclerosus et atrophicus*", which has been broadly used until recently. The term has changed to the contemporary "*lichen sclerosus*", since not all cases exhibit atrophy of the overlying epithelium [2]. The exact cause of this entity remains relatively obscure, and infectious, genetic, traumatic and autoimmune factors may participate in the pathogenesis [3].

LS is considered to be a chronic inflammatory disease affecting mainly the genital mucosa as well as the perianal region and the skin of the neck and the upper trunk [4]. The skin lesions are mainly characterised by flat, polygonal papules that form plaques. Genital lesions present clinically as atrophic plaques, purpuric lesions and dyspareunia in women, whereas phimosis and balanitis xerotica obliterans may occur in males [1]. Extragenital lesions have been reported in 15 - 20% of cases [3].

Although oral lesions are relatively rare, they can be the primary or the only manifestation of the disease [1]. Oral lesions are characterised by ivory or porcelain-white, round plaques. The main affected areas are the lips, buccal mucosa, and tongue and, less frequently, the gingivae and the anterior tonsillar pillar. The atrophic and sclerotic patches are commonly asymptomatic, although pruritus, soreness, burning, discomfort and tightening resulting in restriction of mouth opening have been reported. Telangiectasia may also be observed [4,5]. Histopathological examination is required for the final diagnosis, as the condition cannot be readily distinguished on clinical grounds alone from lichen planus, leukoplakia and localised scleroderma [6]. LS treatment mainly consists of the use of topical corticosteroids, although therapy in asymptomatic cases is not mandatory [2].

Histopathologically, oral LS exhibits atrophic, and sometimes focally thickened, epithelium and subepithelial homogenization and hyalinization, with scarcity or loss of elastic fibres. There is a band-like subepithelial leukocytic inflammatory infiltrate that can move to the deeper portions of the lamina propria, following the maturation of the lesion [1].

The purpose of this study is to present a case of oral lichen sclerosus, diagnosed by utilising the histochemical Shikata's modified orcein stain, as well as a thorough review of all previously published oral lichen sclerosus cases in the English language literature.

CASE DESCRIPTION AND RESULTS

A 32-year-old Caucasian woman presented in an Oral Medicine Private Practice Clinic with white patches on the left upper lip on April 2012. The lesions were initially observed by the patient 3 months ago and were asymptomatic. She had been examined by her dermatologist with a clinical diagnosis of "lichen" or vitiligo. No cutaneous or genital lesions were noticed. No treatment of the oral lesions had been prescribed because of their asymptomatic nature. However, the patient considered that the lesions were cosmetically objectionable. Her medical history was non-contributory.

Clinical examination revealed asymptomatic white patches with irregular borders on the skin, the vermillion border and the mucosa of the left upper lip (Figure 1). In addition, erythematous areas were observed intermingled with the white lesions on the left labial mucosa (Figure 2).

Under local anaesthesia, an incisional biopsy of the mucosal labial lesion was performed, taking a tissue specimen of 1.5 x 0.7 x 0.4 cm in dimension. Five μm thick formalin-fixed and paraffin-embedded tissue sections stained with hematoxylin and eosin were procceded for histopathological examination, which revealed an atrophic stratified squamous epithelium without rete pegs formation. The subepithelial connective tissue was dense, acellular, and hyalinized. Underneath the acellular zone, a moderate band-like lympohistiocytic inflammatory infiltrate was noticed (Figures 3A - C). Tissue sections of same thickness were further evaluated histochemically with Shikata's modified orcein stain (treated with acidified potassium permanganate, decolourized with 1% oxalic acid, and finally stained with Shikata orcein), which revealed distinctive scarcity or loss of elastin fibres in the hyalinized papillary lamina propria (Figure 3D). A final diagnosis of lichen sclerosus was rendered.

Treatment consisted of topical application of fluocinonide 0.05% gel three times per day. Follow-up examination after 2 weeks revealed significant improvement of the lesions (Figures 4 and 5). The patient was placed on long-term follow-up course (3-monthly for 2 years and 6-monthly until now), without any recurrences noticed so far. The patient gave us her permission to use details of the case for publication.

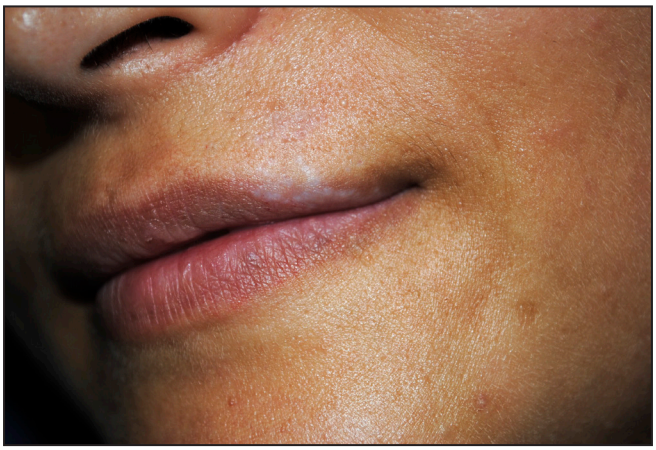

Figure 1. White plaque on the skin and the vermilion border of the left upper lip (x1/2).

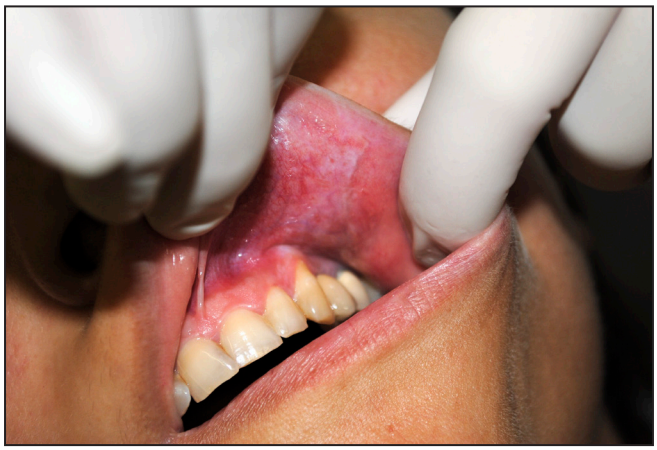

Figure 2. Intermingled white patches and erythematous areas on the mucosa of the upper lip (x1/2).

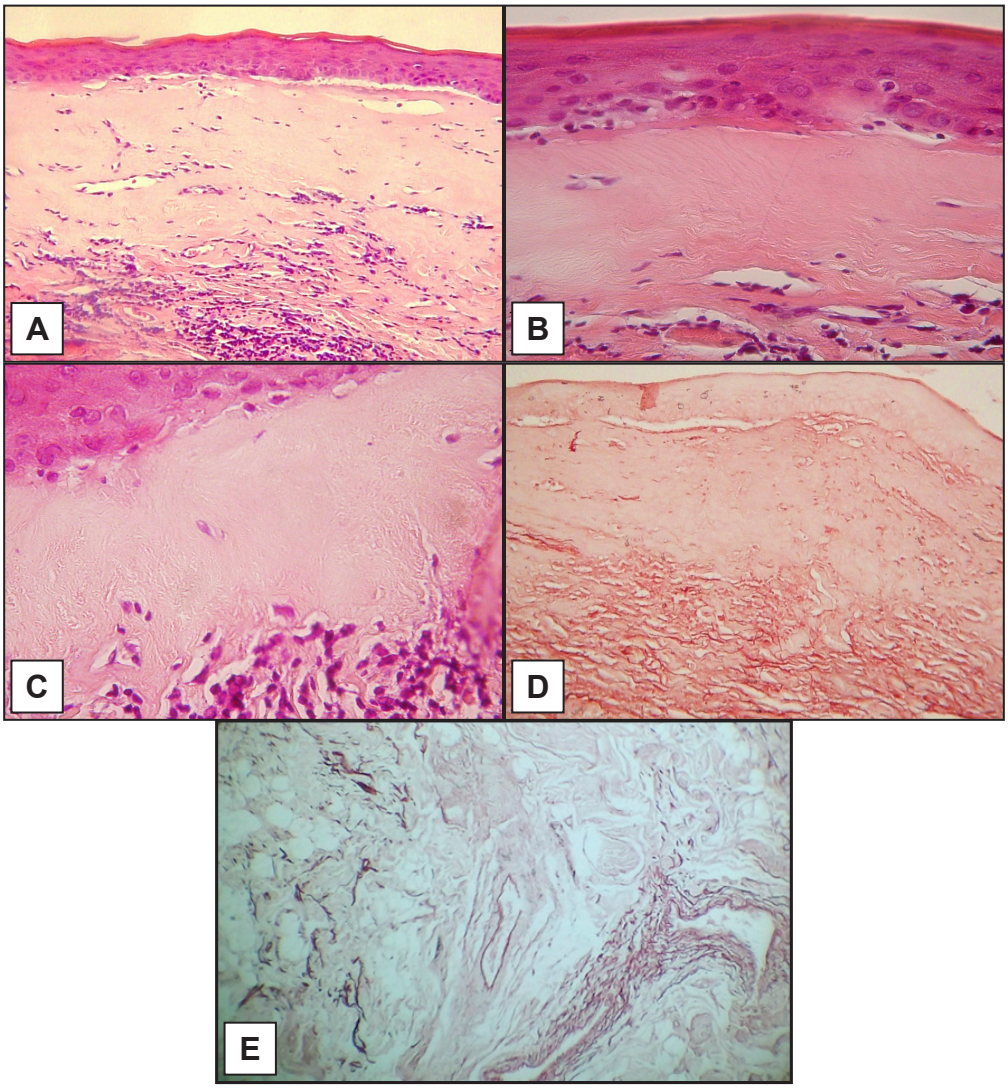

Figure 3. A = histopathologic examination revealed epithelial atrophy, loss of rete pegs, hyalinization of the lamina propria and deep lymphohistiocytic inflammation (hematoxylin and eosin stain, original magnification x100).
B = hyalinization and loss of cellularity of the subepithelial lamina propria (hematoxylin and eosin stain, original magnification x200).
C = hyalinization and loss of cellularity of the subepithelial lamina propria (hematoxylin and eosin stain, original magnification x200).
D = histochemical Shikata's modified orcein stain revealed loss of elastin fibres in the hyalinized subepithelial area (original magnification x100).
E = normal deep connective tissue, stained with Shikata's modified orcein stain, showing plenty of brown-coloured elastin fibres in a perivascular orientation (original magnification x100).

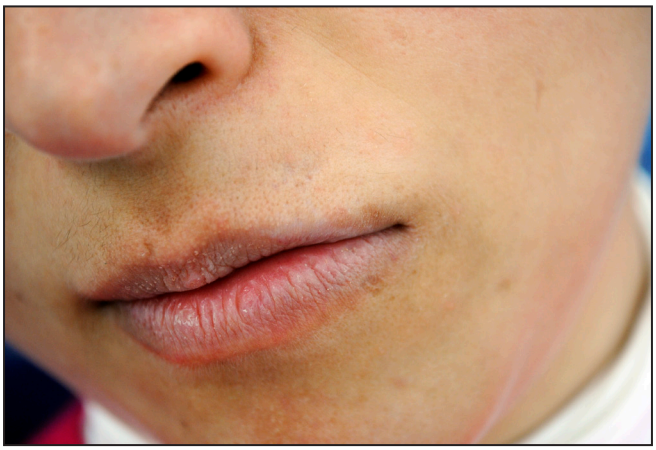

Figure 4. Resolution of the white plaque on the skin and vermillion border after treatment (x1/2).

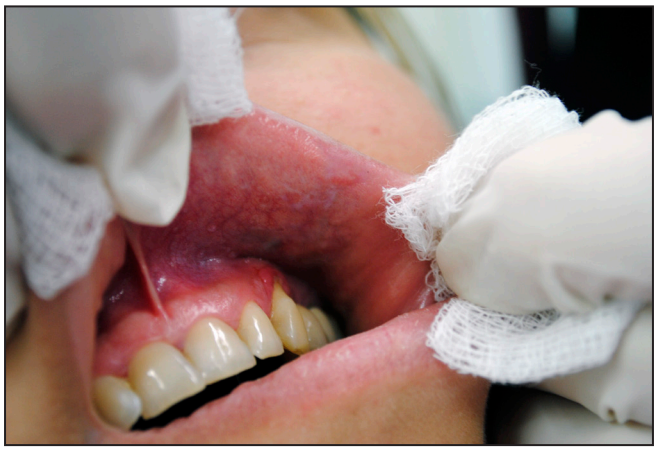

Figure 5. Intermingled white patches and erythematous areas on the mucosa of the upper lip (x1/2).

DISCUSSION

LS is an unusual mucocutaneous disease that mainly affects the anogenital area and the skin. It mainly affects females with varying degrees of incidence (1 - 10:1) [1,3,4]. LS may affect patients of all ages, and in women there is a noticeable peak of occurrence in the second and fifth-sixth decades; men are mainly affected in the fourth-sixth decades, despite the fact that LS has been also described in boys with phimosis [2,3,7].

Oral manifestations of LS are rare and may appear with or without accompanying extraoral lesions [6]. A systematic review of the English language literature was conducted, using searching keywords: "oral lichen sclerosus", "oral white striae", "oral white patches", in MEDLINE, Web of Science, and Scopus databases, taking into consideration any case of histopathologically-assessed LS affecting the oral mucosa, with or without extraoral involvement. Since 1957, when Miller [8] published the first well-documented oral LS case, few cases without skin or genital involvement have been reported. Following data items of the reported oral LS cases were summarized in Table 1: "Author(s)", "Patient's sex/age", "Oral site(s)", "Oral symptoms", "Skin lesions", "Genital lesions", "Duration" (before clinical presentation), "Treatment". Attili and Attili [9] reported also 27 cases of LS of the lips; 20 out of them were the only manifestation of the disease. Our study adds one more case of LS that appeared solely in the oral cavity and perioral tissues, without further mucocutaneous involvement.

A wide range of ages has been recorded on the published cases with oral involvement, ranging from 7 up to 67 years. Most of them were females. Regarding localisation of the lesions, the lower and the upper lip, the buccal mucosa and the tongue were the most common areas of oral LS. The gingiva, the palate, and the anterior tonsillar pillar are also sites of involvement (Table 1) [1,4].

Clinically, oral LS lesions appear as white papules that coalesce to form plaques. Striae and superficial erosions may also be observed; in such cases, LS may be indistinguishable from erosive lichen planus [10]. The lesions are mainly asymptomatic, although symptomatic cases of oral LS have been described (Table 1) [4]. Moreover, Jiménez et al. [11] observed loss of clinical periodontal attachment when the lesions were located on gingiva.

Histopathologically, the affected epithelium in oral LS exhibits similar features to those of lichen planus in various degrees (such as atrophy or hyperplasia and hydropic degeneration of basal cells) [6]. Generally, it is considered that skin lesions are mainly characterised by hyperplasia, whereas atrophic epithelium with loss of rete ridges is observed in mucosal lesions [5,12]. However, the most important features, which are of diagnostic importance, are observed in the connective tissue. The papillary lamina propria is characterised by hyalinization and loss of cellularity [13,14]. Special histochemical techniques, such as Verhoeff-Van Gieson or Shikata's modified orcein stain, show scarcity or loss of elastin fibres, a distinctive and diagnostic histopathological feature of LS [15]. Underneath the acellular area, a band-like lichenoid lymphocytic infiltrate, consisted of CD4+ and CD8+ T-cells is observed, and its location depends on the degree of maturation of the lesion; the more immature the lesion, the more superficial the infiltrate [4]. The histopathological examination is mandatory for confirmation of the diagnosis.

Table 1. Summary of the reported oral LS cases in the English literature

Author(s)	Patient's age/sex	Oral site(s)	Oral symptoms	Skin lesions	Genital lesions	Duration	Treatment
Sherlin et al. [1]	60/M	Gingival and buccal mucosa	No	No	No	6 months	No
Louvain et al. [2]	70/F	Right buccal mucosa, tongue, gingiva	No	Yes	Yes	3 months	Tacrolimus
Azevedo et al. [4]	19/M	Buccal and labial mucosa	No	No	No	8 months	No
	34/F	Upper lip and labial mucosa	Yes	No	No	3 months	Surgical excision
	11/F	Vermillion upper lip and labial mucosa	No	No	Yes	36 months	No
	38/F	Buccal and labial mucosa	Yes	Yes	No	20 days	Triamcinolone/ colchicine
	31/F	Lower labial mucosa and tongue	No	Yes	No	6 months	Surgical excision/ colchicines
	28/F	Tongue, buccal and labial mucosa	Yes	No	No	7 months	No
Jensen et al. [5]	10/F	Buccal mucosa and sulcus	No	No	NR	1 year	No
Jiménez et al. [6]	31/F	Gingiva, buccal sulcus and labial mucosa	No	Yes	No	2 years	Triamcinolone
Walsh et al. [7]	16/M	Vermilion upper lip, labial mucosa	No	Yes	No	9 months	Methotrexate/prednisone
Miller [8]	48/F	Buccal mucosa	No	Yes	Yes	7 months	NR
Kaur et al. [10]	16/M	Upper lip and gingiva	Yes	Yes	No	1 year	Prednisolone and intralesional triamcinolone
Jiménez et al. [11]	19/F	Gingiva and frenulum of upper lip	Yes	No	No	NR	Triamcinolone
Brown et al. [12]	44/M	Soft palate	No	No	No	NR	No
	18/M	Lower lip	No	Yes	No	1 year	Clobetasol/testosterone
Chaudhry et al. [13]	60/F	Dorsum of the tongue	Yes	Yes	Yes	4 years	Benzydamine/ betamethazone
Mendonça et al. [14]	20/F	Vermilion lower lip, labial mucosa	No	No	No	NR	No
Buajeeb et al. [15]	22/F	Buccal mucosa, buccal sulcus vermilion lower lip and labial mucosa	Yes	No	No	NR	Triamcinolone and methylprednisolone
Kim et al. [22]	7/F	Left lower lip and buccal mucosa	No	No	No	2 years	Topical steroids/ pimecrolimus
de Araujo et al. [27]	26/F	Upper labial mucosa, buccal sulcus and gingiva	NR	Yes	No	NR	NR
Siar and Ng [28]	25/M	Buccal mucosa and lips	Yes	Yes	No	6 months	Triamcinolone
Macleod and Soames [29]	57/F	Hard palate and tongue	Yes	No	No	Several months	NR
Schulten et al. [30]	59/F	Upper lip, left commissurae and tongue	No	No	No	3 months	No
	12/M	Left lower lip	No	No	No	9 months	Incisional biopsy
Rajlawat et al. [31]	14/F	Lower lip	No	No	No	1 year	Clobetasone
Kelly et al. [32]	10/F	Vermilion lower lip, labial mucosa	No	No	No	2 years	Clobetasol
Liu et al. [33]	54/F	Tongue dorsum	No	No	No	10 days	Tacrolimus
	58/F	Tongue dorsum	No	No	No	2 years	Tacrolimus
De Aquino Xavier et al. [34]	47/M	Upper maxillary alveolar mucosa	No	No	No	NR	Surgical excision
George et al. [35]	20/M	Upper labial mucosa, adjacent maxillary gingiva	NR	NR	NR	NR	NR
Tupsakhare et al. [36]	67/M	Soft palate	No	No	No	11 years	No
Marangon Júnior et al. [37]	44/M	Upper lip	Yes	No	No	6 years	No
Wakamatsu et al. [38]	54/F	Lower lip	No	No	No	3 years	Tacrolimus
Ravits and Welsh [39][a]	24/M	Buccal mucosa, gingiva and palate	NR	No	No	4 months	Vit A
	42/M	Buccal mucosa and palate	NR	No	Yes	7 years	Vit A, Vit D2, padothenyl alcohol
Sarkany [40]	57/F	Front of hard palate	No	Yes	Yes	10 years	NR
Present case	32/F	Upper lip, vermilion and labial mucosa	No	No	No	3 months	Fluocinonide gel

[a]Ravits and Welsh [39] reported also a third case clinically suggestive of lichen sclerosus, but with no histopathological confirmation.
NR = not reported; M = male; F = female.

The ultrastructural findings of LS include the presence of broad diameter collagen fibres with loss of cross-striations, holes and gaps of the basal epithelial layer, increased number of Langerhan's cells and decreased number of melanocytes [3,4,16]. Immunofluorescent study is not diagnostically contributory, since it usually reveals only fibrinogen deposits at the basement membrane, equally to lichen planus [3,15].

The differential diagnosis includes mainly localised scleroderma (morphea), lichen planus, leukoplakia, vitiligo, and oral submucous fibrosis [6,10,11]. Localised scleroderma can be easily excluded since it is not characterized by neither vacuolar degeneration of basal cells nor the lichenoid inflammatory reaction and loss of elastin fibres [15]. The immature lesions of LS, in which the lichenoid inflammation is more superficial, are quite difficult to be distinguished from lichen planus. This difficulty is easily overcome in more mature lesions, since in LS there is a thick hyalinization of the papillary lamina propria, similar to our case [14]. The same distinctive histopathological features are used in order to exclude leukoplakia and oral submucous fibrosis, along with the lack of dysplastic features in LS. Despite their decreased number, the presence of melanocytes (which are S-100 positive) in LS lesions is evident, excluding vitiligo [14]. Nonetheless, it should be noted that cases with co-existed lichen sclerosus, lichen planus and localised scleroderma have been reported to occur concurrently at the same patient [17].

The pathogenesis of LS remains obscure and several factors have been implicated. An immune-mediated disorder is the most plausible explanation [3]. There are studies that describe the co-existence of LS with autoimmunologic disorders, such as thyroiditis, diabetes mellitus, alopecia areata, pernicious anaemia or vitiligo [6]. Whether these entities share common pathogenetic mechanisms or its concurrence is an accidental finding is still unknown. Moreover, autoantibodies against the glycoprotein Extracellular Matrix Protein 1 (ECM1) have been detected in the serum of patients affected by LS [1].

ECM1 is a molecule that plays various roles; within the epithelium, it is correlated with the control of the keratinocyte differentiation, while it modulates the organisation of the connective tissue as one of the major binding partners to perlecan [1,18]. Lipoid proteinosis, a rare metabolic disorder with uncommon oral manifestations similar to LS, is characterised by mutations in ECM1 gene [18]. Thus, it may be hypothesized that a dysfunction of ECM1 may produce the histopathological alterations that are observed in LS cases. Interleukin-6 (IL-6) has also been suggested to play an important role in the pathogenesis of LS, since an increased amount of this cytokine was detected in the atrophic epithelium of LS cases [3].

Furthermore, there are studies that suggest a genetic predisposition. LS has been observed within members of the same family (familial LS) or in identical twins [3,19]. There is no specific association of LS with HLA antigens, though studies suggest a possible correlation with HLA-DQ7 alleles [20]. The possible connection of LS with particular microbial infections, especially by *Borrelia burgdorferi*, has also been mentioned, as 60% of LS specimens revealed superficial *Borrelia* colonisation [4]. In many cases, significant clinical improvement was noticed after antibiotic treatment. Viral infections, especially by HPV, have been reported to cause or contribute to the development of LS lesions, especially in genitalia; this correlation though remains questionable [21]. Finally, local irritation or traumas have been linked to the pathogenesis of LS, especially for cutaneous and genital lesions. In these cases, the Köebner phenomenon (occurrence of LS in areas of already damaged skin or mucosa) has been suggested as the potential pathway [3].

Until now, there is no specific successful treatment for the oral manifestation of LS. Treatment is commonly provided in cases of LS with soreness and discomfort or in cases that cosmetic concerns have arisen. Due to the proposed immune-mediated pathogenesis of LS, along with the inflammatory component observed clinically and microscopically, the most commonly used treatment modalities involve topical application or intralesional injections of corticosteroids and topical calcineurin inhibitors (tacrolimus, pimecrolimus); cryotherapy, and surgical excision for small lesions have also been utilised (Table 1) [1,2,22]. Colchicine, a drug that is mainly used in other mucocutaeous diseases, like Behçet's syndrome, psoriasis and scleroderma, has also been used in a limited number of LS cases [4]. For the genital manifestations, topical application of testosterone, narrow-band UVB and psoralen UVA (PUVA) are proposed to be effective in reduction of the lesions following one or more courses [23,24].

In the literature, no malignant transformation of oral LS cases has been reported. Nevertheless, genital LS lesions have been associated with a 3 - 5% relative risk for the development of squamous cell carcinoma [3]. Since oral LS may be accompanied by genital lesions, it is highly recommended that the patient should undergo a thorough clinical examination and be placed in a long-term follow-up course [24-26].

CONCLUSIONS

The pertinent English language literature review has revealed a few cases of oral lichen sclerosus [1,2,4-15,22,27-40]. Although considerably uncommon, oral manifestations of lichen sclerosus should be included in the clinical differential diagnosis of white patches of the oral mucosa and the vermillion border of the lips. Further histopathological evaluation of the specimen with Shikata's modified orcein stain is useful for rendering a proper diagnosis.

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The authors report no acknowledgments and no conflicts of interest related to this study.

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