Lichen sclerosus of the oral mucosa: A case report

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Abstract
Lichen sclerosus or lichen sclerosus et atrophicus is a chronic inflammatory disease predominantly affecting the genital mucosa and skin. Clinically, it is characterized by white atrophic plaques in the anogenital region. The lesions are generally asymptomatic, but may cause discomfort with itching and pain. Extragenital mucosal involvement is very unusual, and lesions limited to the oral mucosa are even less frequent. Knowledge of such lesions is important in order to establish a differential diagnosis with other white oral lesions, and histological confirmation is required. We present the case of a 31-year-old woman with a well delimited, pearly white lesion located in the upper gingival mucosa, lip mucosa and adjacent skin. The lesion had led to loss of periodontal attachment of the affected tooth (2.3), causing pain in response to tooth brushing. The biopsy confirmed lichen sclerosus, and treatment was provided in the form of intralesional corticoid injections, followed by improvement of the mucosal lesion, though without recovery of the periodontal loss.

Key words: *Lichen sclerosus, scleroatrophic lichen, oral mucosa, periodontal loss.*

Introduction
Lichen sclerosus or scleroatrophic lichen is a chronic inflammatory disease most often seen in women, though men and children can also be affected. The lesions are most commonly found on the skin and anogenital region (affected in 80% of cases), and are characterized by white atrophic maculae. The lesions are generally asymptomatic, but may cause discomfort with itching, bleeding and functional impairment. At skin level, the lesions manifest as isolated, non-elevated ivory white maculae that tend towards confluence (1-4). Involvement of the extragenital mucosas is very uncommon, though there have been reports of lesions in other mucosas, as well as of isolated oral mucosal involvement - without genital or skin lesions. The low frequency of the disease and the absence of symptoms sometimes causes the lesions to go unnoticed; as a result, they can spread and lead to tooth loss when the gingival tissue is affected. Knowledge of such lesions is therefore important in order to identify them and establish a differential diagnosis with other white oral lesions. Histological confirmation of the diagnosis is required.

Case report
A 31-year-old woman without disease antecedents of interest or known allergies or toxic habits (smoking, alcohol abuse) reported to the Service of Stomatology (Valencia University General Hospital, Valencia, Spain) with a whitish oral lesion that had been present for the previous two years and caused discomfort on tooth brushing.
Clinical examination revealed a non-elevated, pearly white homogeneous macula with well defined limits, extending from the vestibular gingival zone of tooth 2.1 to 2.5, and to

the mucosa and skin of the upper lip. The lesion was asymptomatic and produced gingival retraction at tooth 2.3, with periodontal loss, causing discomfort on brushing. Palpation yielded a solid sensation, and the resulting suspected clinical diagnosis was vitiligo or lichen sclerosus. (Fig. 1, 2).

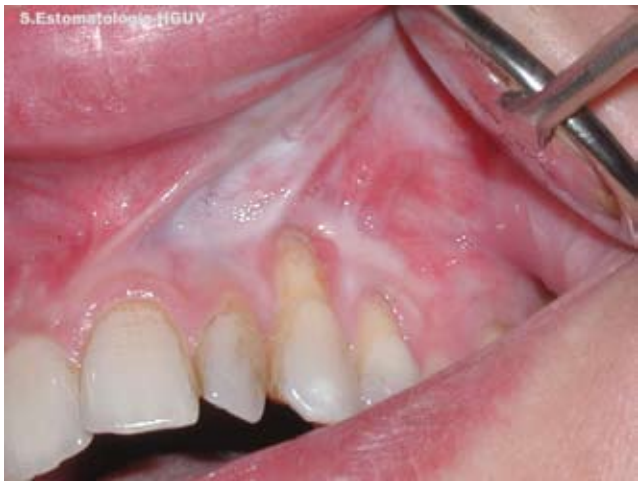

Fig. 1. Clinical view of the oral lesion.

Fig. 2. Clinical view of the skin lesion.

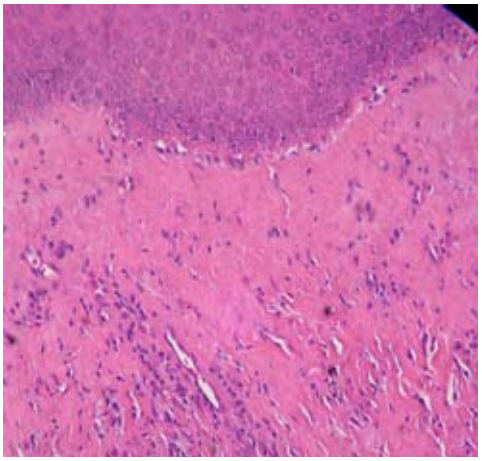

Fig. 3. Histopathological view of the oral lesion.

Laboratory tests were requested, showing normal results for erythrocyte sedimentation rate and negative circulating antibodies (ANA, SSA, SSB, Sm, RNP, histones, DNA, RF).

An incisional biopsy of the lip mucosa was obtained. Following hematoxylin-eosin staining, the histological study revealed an epithelium with normal keratinization and atrophy of the papillary crests. The connective tissue showed collagen fiber homogenization with hyalinization of the lamina propria due to the accumulation of an amorphous eosinophilic material forming loop-like structures, displacing a lymphocytic inflammatory infiltrate towards deeper layers, and located at the periphery of dilated vascular structures. (Fig. 3).

There were no skin lesions, and the patient was referred to the Service of Gynecology where the presence of genital lesions was discarded.

Treatment was provided in the form of perilesional 40-mg triamcinolone infiltrations (Trigon depot®) (7 infiltrations: one every 10 days), followed by improvement of the mucosal lesion, with persistence of the gingival attachment loss. (Fig. 4).

The patient is presently subjected to periodic controls, and no new oral, genital or skin lesions have appeared.

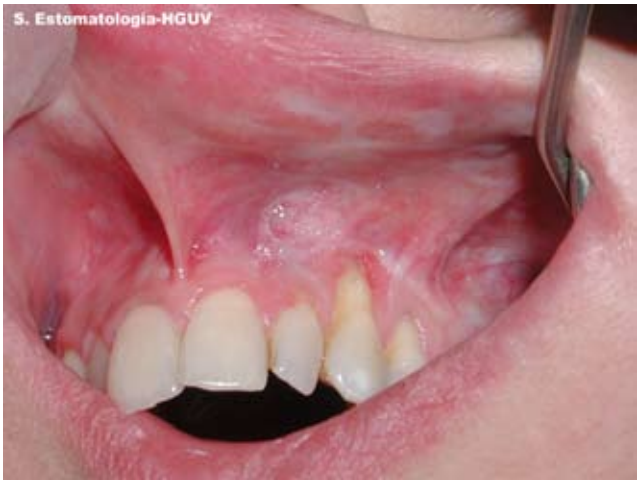

Fig. 4. Clinical view of the skin lesion after treatment.

Discussion

Lichen sclerosus (LC) is a mucocutaneous chronic inflammatory disease of uncertain origin, clinically characterized by the presence of well delimited white maculae, located fundamentally on the skin and genitals, and very rarely in the oral cavity (1-4). The prevalence of LS located only on the oral mucosa or on the oral mucosa and adjacent skin is extremely low (1-13)(Table 1). This fact, and the lack of symptoms, may often cause the lesions to go unnoticed by the dental professional.

The etiology is not known, though the condition has been related to autoimmune disorders - some studies having

Table 1. Lichen sclerosus et atrophicus confirmed by biopsy, with exclusive involvement of the oral mucosa and adjacent skin.

Author	Year	Location	Sex/age	Skin*
Ravits(1)	1957	oral mucosa, gums	M/24	no
Araújo et al. (2)	1985	lip mucosa, gums	F/26	subnasal
Macleod and Soames (3)	1991	tongue and palate	F/57	no
Schulten et al. (4)	1993	- lip, tongue - lip mucosa	F/59 M/12	no no
Brown et al. (5)	1997	- lip mucosa - soft palate	M/18 M/44	lip no
Buajeeb et al. (6)	1999	gums, cheek and lip mucosa	F/22	no
Jiménez et al. (7)	2002	lip mucosa, gums	F/19	no
Jensen et al. (8)	2002	cheek mucosa	F/10	no
Kaur et al. (9)	2002	lip mucosa, gums,	M/16	nose, lip
Mendonca et al. (10)	2004	lip	F/20	no
Rajlawat et al. (11)	2004	lip	F/14	no
Kelly et al. (12)	2006	lip	F/10	no
Chaudhry et al. (13)	2006	tongue		no

reported an association with autoimmune diseases such as psoriasis and thyroid autoimmune processes (14). A genetic predisposition has been described, based on the observation of an increased familial prevalence. In this context, the disease has been associated with certain HLA antigens - fundamentally DQ7. Our patient reported no autoimmune pathology or similar lesions in any relative.

The oral lesions manifest as well delimited, pearly white and non-elevated, level maculae of variable size (from several millimeters to extensive lesions). The lesions are usually asymptomatic and can be located anywhere on the oral mucosa. The lesions appear indurated in response to palpation. There also have been reports of telangiectasia in relation to the lesions (8).

A gingival location of these lesions leads to periodontal attachment loss as seen both in our patient and in other cases reported in the literature (2,6,7,9). This often causes tooth loss, but constitutes a problem strictly confined to the area of the lesion - not generalized as in the case of scleroderma for example.

The absence of reticular streaks must be stressed, both at the periphery of the lesions and in other locations - thus allowing clinical differential diagnosis with lichen planus. Other differential diagnoses that must be taken into consideration are leukoplakia, oral submucosal fibrosis, vitiligo and scleroderma. A histological study is required to differentiate these conditions.

In 1957, Ravitis (1) was the first to describe the microscopic features of these lesions located in the oral cavity - the findings being quite similar to those of the skin lesions, with the exception of hyperkeratosis (10). The microscopic study reveals epithelial atrophy, local hydropic degeneration of the basal cells, loss of the epithelial crests, and homogenization of the underlying connective tissue with a reduction in elastic fiber presence, leading to hyalinization and connective tissue sclerosis. The initial lesions show a mononuclear infiltrate of the sub-basal layer that posteriorly displaces towards deeper layers in the region of the vascular plexus, in more mature lesions (9).

These characteristics allow us to differentiate the lesions from lichen planus, where a band-like subepithelial lymphocytic infiltrate is seen, and from localized scleroderma, where the changes are seen in the deep layers of the dermis - with increased collagen production and no reduction or loss of elastic fibers (7,9). The presence of melanocytes among the basal cells differentiates LS from vitiligo, while the absence of vascular lumen obliteration or reduction distinguishes the condition from submucosal fibrosis (10).

The most widely used treatment is local corticoids in the form of clobetasol propionate ointment at genital level or as intralesional injections of triamcinolone at oral level - resulting in improvement of the mucosal lesions, as in our patient. The anogenital lesions also have been treated with 0.1% tacrolimus ointment (15).

Corticoid therapy is unable to improve periodontal involvement, and in some cases extraction of the affected tooth is required due to mobility. Some authors recommend no treatment of the oral lesions, due to the absence of symptoms and their benign behavior, provided there are no adverse esthetic effects.

Although there have been no reports of malignant trans-

formation of LS, the lesions have been associated with squamous cell carcinoma lesions of the genital mucosa, but not in relation to extragenital lesions or children. The patients should be subjected to follow-up, to control the possible appearance of new oral, genital or skin lesions.

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