Up-to Date Review And Case Report

Isolated lichen planus of the lips: cases reports and literature review

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Abstract – Introduction: Lichen planus is an inflammatory mucocutaneous dermatosis involving skin, appendages and mucosa. Oral mucosa is the most commonly involved in all its sites, rarely the lips especially when isolated. The aim was to conduct a literature review about isolated lichen planus of the lips and reporting two case reports of this lesion in order to highlight epidemiologic, clinical and histological features and therapeutic modalities of this lesion. **Observations:** Case report 1: a 34-year-old diabetic male patient consulted for an erosive, crusted and hemorrhagic cheilitis of the lower lip. Clinical and histological examination led to the diagnosis of isolated lichen planus of the lips. Case report 2: a 33-year-old female patient was referred from dermatology department for biopsy of chronic cheilitis of the lower lip. Clinical and histological examination confirmed the diagnosis of isolated lichen planus of the lips. **Discussion:** The review based on 34 case reports of isolated lichen planus of the lips, in addition to literature data confirmed that it is a benign rare lesion affecting mostly male patients having middle age with preponderance of the lower lip, its erosive form is the most frequent and it presents a favorable healing with topical treatment particularly corticosteroids.

Introduction

Oral lichen planus is a benign inflammatory dermatosis which may involve all sites of oral mucosa. It occurs mostly on the buccal mucosa, tongue, gingiva and palate. Lip involvement, particularly if isolated, is unusual.

Isolated lichen planus of the lips is underreported in the literature. In addition, its clinical features are usually confusing leading to many wrong diagnoses. That's motivated this article.

The aim was to conduct a literature review about isolated lichen planus of the lips and reporting two case reports of this lesion, in order to investigate its epidemiologic, clinical and histological features, besides to the therapeutic modalities.

Case report no. 1

A 34-year-old diabetic male patient presented with a 3-month history of erosive crusted and hemorrhagic cheilitis of the lower lip.

The extra oral examination showed ulceration of 3 cm of diameter with marked crusted and bleeding areas. Perilesional white keratotic striae were also revealed (Fig. 1).

The upper lip and oral mucosa were normal.

Anamnesis and physical examination revealed no history of previous skin disorder, local trauma, excessive sun exposure or recent drug intake. Therefore, actinic cheilitis, allergic contact cheilitis, Stevens-Johnson syndrome were ruled out.

Biopsy and direct immunofluorescence were performed. Direct immunofluorescence was negative.

Histological examination showed hyperplastic epithelioma, a parakeratosis, liquefaction and degeneration of the basal layer which was irregular, in addition to a band-like plasmocytes infiltrate in the dermal-epidermal interface (Fig. 2).

These features were consistent with oral lichen planus of the lower lip.

The patient was treated with topical corticosteroids: Clobetasol twice a day.

Within five months the lesion had entirely resolved (Fig. 3).

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Fig. 1. Crusted hemorrhagic ulceration along the lower lip with lacy white streaks on the lateral border of the lesion.

Fig. 2. HE * 40: Hyperplastic epithelioma, parakeratosis, liquefaction and degeneration of the basal layer. A band-like plasmacytes infiltrate in the dermal-epidermal interface.

Case report no. 2

A 33-year-old female patient had been referred from dermatology department for biopsy of chronic cheilitis of the lower lip that had evaluated for 8 years.

Fig. 3. Complete resolution within five months, persistence of atrophic and reticular aspect of the lower lip.

Fig. 4. Swelling and atrophic mucosa of the lower lip with erosion, fissures and crusted areas. Lesions are surrounded by lacy white reticular streaks.

Previous biopsy and direct immunofluorescence of the lower lip were inconclusive.

Her familial and post medical history was non-contributory.

At anamnesis, she reported recurrence of edema and crusting of the lower lip with hemorrhagic fissures. She had no history of drug intake, local injury and had not experienced excessive sun exposure.

Exobuccal examination revealed swelling, atrophic mucosa and erosion of the lower lip with fissures and crusted hemorrhagic areas, in addition to lacy white streaks forming a reticular pattern in the lateral border of the lesion (Fig. 4),

Fig. 5. HE*40: Apoptotic bodies in the epithelium, parakeratosis and a dense band-like lymphocytic infiltrate in the connective tissue with a degeneration of the basal layer.

Fig. 6. Entirely resolved within seven months.

therefore a provisional diagnosis of oral lichen planus was made.

The upper lip was uninvolved. The remainder of the mucosal surfaces and the skin showed no abnormality. Direct immunofluorescence was negative. Biopsy from the lower lip showed an epithelium full of apoptotic bodies with parakeratosis and a dense band-like lymphocytic infiltrate in the connective tissue with a degeneration of the basal layer (Fig. 5).

Treatment was commenced with a topical corticosteroid: betamethasone once a day at night. The patient showed a favorable improvement, but after three months, edema and crusting were revealed in the lower lip. So, she was recommended a sun screen and the application of topical fluocinonide for two weeks twice a day. Within three months the lesion was entirely resolved again (Fig. 6).

Discussion

A review of the literature was conducted on the database Medline via its interface PubMed using Mesh Keywords : "lip", "lichen planus, oral", "therapeutics" and combining the following Boolean equations : "lip" and "lichen planus, oral" / "lip" and "lichen planus, oral" and "therapeutics", in the period from 1939 to 2019. This bibliographic research concluded to 32 case reports about isolated lichen planus of the lips from 19 articles.

The parameters extracted from these cases were summarized in Table I and they concerned: age, sex, clinical form, localization, skin involvement, systemic pathologies, date of appearance, treatment and outcomes (Tab. I).

Lichen planus is an inflammatory benign condition of the skin and mucosa whose etiology is still unknown.

In the oral cavity, commonly involved sites are buccal mucosa and tongue. However, gingiva, floor of mouth, palate and lips are rarely affected [1,2].

In fact, the prevalence of lip involvement varies from 6.3 to 29.4% [1].

Isolated lip lichen planus is less frequent, its prevalence reported in the literature varies from 0.51 to 8.9% [1].

Isolated lichen planus of the lips involves patients in the middle age with a male preponderance [1]. This has been confirmed by our review. In fact, the results showed 24 males/10 females and 27 patients were aged between 40 and 74 years (Tab. I).

Due to their anatomic localization, lips are currently subject to many injuries: such as sun exposure, make up application, biting. Therefore, clinical features of isolated oral lichen planus of the lips are not pathognomonic and usually misdiagnosed and may mimic many other types of cheilitis.

Isolated lichen planus of the lips appears as whitish, reddish or mixed surfaces with crusting, erosion and ulceration that may be associated to some blisters along the vermillion of the upper, lower or the two lips [1]. These features were identified in our two patients.

All clinical variants of oral lichen planus that have been described in the literature may also be identified in the isolated form of the lips which are: reticular, papular, erosive, bullous and atrophic form [1,3].

The reticular form is the most typical characterized by Wickham's striae [1]. However, the erosive one is the most common according to the literature [1,4]. In our review, the erosive form was the frequent one (22 cases), then the hyperkeratotic or plaque-like form (5 cases), the reticular one was presented in four cases and finally the less common was the annular form (2 cases) (Tab. I).

Lower lip involvement shows a clear predominance compared to the upper lip. In fact, the lower/upper lip involvement ratio was 6:5 [1]. These data were also concluded from our review which revealed a lower lip involvement in 27 cases, five cases of upper and lower lip involvement and only one case of upper lip involvement (Tab. I).

Table I. Clinical cases about isolated lichen planus of the lips reported in the literature from 1939 to 2019.

No. of case	Article and year	Age	Sex	Localization	Clinical form	Date of appearance	Skin involvement	Systemic pathologies	Treatment	Evolution with treatment
1	Whittle CH, 1939 ¹³	69	Male	Lower lip	Plaque	-	Genital mucosa	No	Mercur, arsenic, X-ray	Stable
2	P. H. ITIN, 1995 ⁵	44	Male	Lower lip	Erosive	3 years	no	No	Acitretin : neotigazon® 30 mg /day prednisone 15 mg /day, sun screen. Betamethasone valerate® cream 0.1% / 2 weeks Chloroquine phosphate 25 mg/day Betamethasone Dipropionate 0.5% ointment	Complete remission in 10 weeks
3	S. ALA.MN, 1996 ⁶	51	Male	Lower lip	Reticular	9 months	no	No		Complete remission in 3 months
4	D De Argila, 1997 ¹¹	51	Male	Lower lip	Erosive	11 years	no	No		Complete remission of symptoms in 6 months
5	Roberto Cecchi, 2002 ¹⁰	43	Male	Lower lip	Reticular	7 months	no	No		Complete remission in 1 months, no symptoms in 4 months
6	Chiang CT, 2002 ¹²	36	Female	Lower lip	Erosive	-	no	No	Prednisolone	Remission
7	Yu Tc, 2003 ¹⁷	44	Male	Lower lip	Erosive	-	no	Hypertension	Globetazol	Remission
8	Donovan JC, 2005 ¹⁸	51	Male	-	Erosive	-	no	HCV-Hepatitis	Tacrolimus	Stable
9	Petruzzi M, 2007 ⁴	52	Female	Lower lip	Hyperkeratotic	6 months	-	No	Globetazol propionate 0.05 % : cream 2/day, Tocopherol oil	Complete remission
10	Petruzzi M, 2007 ⁴	54	Male	Lower lip	Erosive, atrophic	10 months	-	No	Globetazol propionate 0.05 % : cream 2/day, Tocopherol oil	Complete remission
11	Petruzzi M, 2007 ⁴	73	Male	Lower lip	Erosive, atrophic	8 months	-	HCV-Hepatitis	Globetazol propionate 0.05 % : cream 2/day, Tocopherol oil	Partial remission
12	Petruzzi M, 2007 ⁴	49	Male	Lower lip	Erosive atrophic	4 months	-	No	Globetazol propionate 0.05 % : cream 2/day, Tocopherol oil	Complete remission
13	Petruzzi M, 2007 ⁴	52	Male	Lower lip	Hyperkeratotic	2 months	-	No	Globetazol propionate 0.05 % : cream 2/day, Tocopherol oil	Complete remission
14	Petruzzi M, 2007 ⁴	62	Male	Lower lip	Atrophic, erosive	8 months	-	Diabetes	Globetazol propionate 0.05 % : cream 2/day, Tocopherol oil	Partial remission
15	Petruzzi M, 2007 ⁴	74	Female	Upper lip / Lower lip	Atrophic, erosive	10 months	-	HCV-Hepatitis	Globetazol propionate 0.05 % : cream 2/day, Tocopherol oil	Complete remission
16	Petruzzi M, 2007 ⁴	60	Male	Lower lip	Atrophic, erosive	3 months	-	HCV-Hepatitis	Globetazol propionate 0.05 % : cream 2/day, Tocopherol oil	Complete remission
17	Petruzzi M, 2007 ⁴	71	Male	Upper lip / Lower lip	Hyperkeratotic	4 months	-	HCV-Hepatitis	Globetazol propionate 0.05 % : cream 2/day, Tocopherol oil	Complete remission
18	Petruzzi M, 2007 ⁴	80	Female	Upper lip / Lower lip	Atrophic, erosive	6 months	-	HCV-Hepatitis	Globetazol propionate 0.05 % : cream 2/day, Tocopherol oil	Complete remission
19	Johnson H, 2008 ¹⁹	42	Female	Lower lip	Erosive	-	no	No	Tacrolimus	Stable
20	Gencoglan G, 2011 ¹⁵	56	Male	Lower lip	Erosive	2 months	no	No	Imiquimod cream 5%, 2/day	Complete remission, no recurrence in 18 months
21	Gencoglan G, 2011 ¹⁵	61	Male	Lower lip	Erosive	6 years	no	No	Imiquimod cream 5%, 2/day	Amelioration within 2 weeks, recurrence within 6 months
22	Gencoglan G, 2011 ¹⁵	65	Male	Lower lip	Reticular	11 years	no	No	Imiquimod cream 5%, 2/day	Remission within 2 weeks
23	Gencoglan G, 2011 ¹⁵	22	Male	Lower lip	Reticular	4 years	no	No	Imiquimod cream 5%, 2/day	Healing in 2 weeks, no recurrence in 5 months
24	Sarika Holmukhe DNB, 2012 ⁸	40	Male	Lower lip	Annular	3 months	no	No	Tacrolimus 0.03 cream 3/day	Remission
25	Domingues E, 2012 ²⁰	44	Male	Lower lip	Erosive	-	yes	No	Globetazol	Regression of the lesion
26	Sugashima Y, 2012 ²¹	32	Female	Upper lip / Lower lip	Annular	-	no	Allergy to zinc	Tacrolimus	

Table I. (continued).

No. of case	Article and year	Age	Sex	Localization	Clinical form	Date of appearance	Skin involvement	Systemic pathologies	Treatment	Evolution with treatment
27	Dillip Kumar Samal, 2015 ³	52	Male	Lower lip	Plaque	1 years	no	No	Biopsy exeresis	No recurrence
28	Nuzzolo P, 2016 ¹	72	Male	Upper lip	Erosive	-	no	Liver insufficiency HCV-hepatitis	Surgical excision	Complete remission
29	Nuzzolo P, 2016 ¹	74	Male	Upper lip / Lower lip	Erosive	-	no	No	Cortisone	Stable
30	Ellie Choi, 2017 ⁹	62	Female	Lower lip	Erosive	1 years	no	No	Tacrolimus hydrocortisone vaseline, sun screen	Complete remission, No recurrence in 2 years
31	FeiYan Yu, 2018 ¹⁶	38	Female	Lower lip	Erosive	10 years	no	No	TCM : "Qingwen Jiedu Kouyankang granules," glucosides, antiinflammatory hormones/4 months	Amelioration in 1 month, no recurrence in 5 months
32	Mathur M, 2019 ²	44	Male	Lower lip	-	4 months	no	No	Betamethasone Dipropionate 0,5 %	Amelioration in 15 days
33	Case report no. 1	34	Male	Lower lip	Erosive	3 months	no	Diabetes	Clobetasol	Complete remission in 5 months
34	Case report no. 2	33	Female	Lower lip	Erosive	8 years	no	No	Betamethasone	Remission in 7 months

Usually lips lesions are symptomatic, mostly when it consists on the erosive variant. Symptoms are dominated by burning, tenderness and tingle sensations with discomfort that are aggravated with spicy and acidic foods.

Unsightly appearance of lip lesions leads to psychological distress reported by some patients [4].

Concomitant cutaneous lesions are exceptional in the genital region. In the review, one case of skin involvement was revealed in the genital lesion (Tab. I).

Histologically, this lesion showed the pathognomonic characters of oral lichen planus which are irregular acanthosis, orthokeratosis with liquefactive vacuolar degeneration of the basal cell layer. In addition, we revealed hypergranulosis, edema and a dense band like lymphocytic infiltrate in the dermal-epidermal interface. Colloid bodies representing necrotic keratinocytes known as Civatte bodies are also identified [1,3,5,6].

Oral lichen planus is a benign dermatosis, nevertheless, some cases of transformation of lichen planus of the lips into squamous cell carcinoma were documented [2-4].

In fact, malignant transformation is still discussed. According to the literature, the rate of this transformation varied from 0.4 to 5.6% [7]. This variation is due to the diversity of clinical forms of oral lichen planus, the difficulty of distinction between this lesion and lichenoid one and other pathologies, besides to the variety of risk factors [7].

Many diagnoses should be ruled out in case of isolated lip lichen planus. For the erosive form it must be differentiated from caustic or traumatic cheilitis, autoimmune blisters dermatosis, erythema multiform, Stevens-Johnson syndrome, herpes or bacterial infection.

For the keratotic variety, the differentiation between leukokeratosis, lupus, graft versus host disease and isolated lip lichen planus may be difficult. Also, actinic cheilitis, atopic dermatosis or some neoplasia must be eliminated [4,5,8,9,10].

The pathogenesis of oral lichen planus is still not completely understood. The auto immune mechanism is the most involved [7]. Some risk factors are reported, such as solar exposure, tobacco and alcohol consumption, mechanic trauma and cosmetic products application. This may explain the greater incidence in the lower lip involvement [4,8,9].

This lesion can be associated with some systemic diseases like hepatitis infection, diabetes, thyroid disorders, Good syndrome, thymoma, graft versus host disease, hypertension [1,7], therefore some laboratory tests are required: HCV serology, diabetes and thyroid function tests. In our review, ten patients presented systemic pathologies: two patients had diabetes, one patient had hypertension and seven had HCV-hepatitis (Tab. I).

Usually, isolated lip lichen planus shows a great remission with topical treatment, most commonly with topical corticosteroids. Systemic and intralesional administration are rarely used [2-4].

Topical steroids such as Clobetasol propionate, Fluticasone propionate are the first line treatment [3]. Also, Betamethasone valerate 0.1%, Betamethasone dipropionate 0.05%, Fluocinonide and Chloroquine phosphate are applicated [2,4,10,11].

Prednisolone is rarely used due to its galenic form that can't be adapted to the labial application [12]. The surgical excision is described in the literature [1,3]. Immunomodulatory agents in form of tacrolimus and cyclosporine are used topically in patients not responding to topical steroids [3], in addition to retinoids alone or in association with corticosteroids [4].

Some other therapeutics are described in the literature: Wittle [13] proposed the treatment with Mercure, Arsenic and X-rays.

Dillenbug described as treatment the laser [14].

Gencoglan proposed the Imiquimod cream 5% [15] and finally in 2018, Feiyan [16] proposed a traditional Chinese medicine comprising "Qingwen Jiedu Kouyarkang granules", total Paeonia glucosides and a combination of hormones and anti-inflammatory agents.

The exploration of the data review confirmed the topical treatment efficacy. The most common treatment used was the Clobetasol which was used in 13 cases with complete remission in 11 patients, then the Tacrolimus (5 cases), the Betamethasone (4 cases), and finally the Imiquimod (4 cases) (Tab. I).

Conclusion

Through this literature review we can conclude that isolated lichen planus of the lips affects preferentially males in the middle age with a lower lip preponderance. The erosive form was the frequent one. This lesion presents a great response to the topical treatment specially corticosteroids.

The prevention by risk factor elimination and oral hygiene maintenance is required to rule out active recurrence. Also, perfect monitoring of eventual cutaneous lesions or other oral localizations is quite necessary in their early diagnosis and treatment and in early detection of possible malignant transformation.

Conflicts of interest: The authors declare that they have no conflicts of interest in relation to this article.

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