

Up-to Date Review And Case Report

Isolated lip involvement in psoriasis: an uncommon aspect of a common dermatologic condition

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Abstract – Introduction: Psoriasis is a chronic inflammatory skin disease that typically affects the extremities, trunk, scalp, and nails. Psoriatic cheilitis as an exclusive presentation is very rare and to our knowledge, only 6 cases have been reported to date. The absence of cutaneous lesions causes diagnostic difficulties that can result in misdiagnosis and inadequate treatment. **Observation:** We reported the case of a 21-year-old woman with a seven years history of scaly plaques of the vermillion of the lips as the only disease manifestation. Her cheilitis was associated with significant psychiatric morbidity. Oral biopsy showed a psoriasiform pattern. Local applications of betamethasone was proposed. **Discussion:** Although lip psoriasis is extremely rare, it can be the sole presentation of psoriasis even in the absence of accompanying skin lesions, other oral manifestations or a family history of psoriasis. **Conclusion:** Lip psoriasis should be considered in the differential diagnosis of chronic or recurrent treatment-resistant labial lesions.

Introduction

Psoriasis is a common disease encountered in dermatology practices. Despite the often widespread nature of this condition, lesions affecting the lips and oral mucosa are uncommon. Fissured tongue and geographic tongue are the most common clinical presentations of oral psoriasis. Psoriasis involvement of the lips is a very rare presentation, with only a handful of cases reported in the literature. Lip involvement can be associated with other cutaneous and/or oral lesions of psoriasis, or can be the sole presentation. This latter case proves to be extremely rare in the literature [1]. We report a rare case of isolated lip psoriasis in a 21-year-old woman evolving approximately 7 years with unusual features and that could be source of misdiagnosis. We also discuss the difficulties in making a definitive diagnosis of oral psoriasis based upon clinical and histological evidence only.

Case report

A 21-year-old woman was referred to our department for further assessment of seven years history of recurrent erosive

cheilitis. She attributed the onset of her disease to exuberant rubbing of the lips during an exam session secondary to an unusual burning and tingling sensation. There was no other identifiable antecedent event or medical or environmental exposure. Within 2 weeks, she developed diffuse yellow-white scale affecting the entirety of both lips. Since that episode, she complained of pain, a burning sensation on her lips and severe discomfort during eating salty and acid food. Lip stick was also irritant.

She initially attempted to treat this with self-debridement and moisturization, to no effect. She was then evaluated by multiple physicians, who prescribed a variety of oral and topical steroids, anti fungals, and anti virals, all to minimal or no effect.

The patient underwent an incisional biopsy of the lower lip 5 years ago that was interpreted as non-specific cheilitis. The diagnosis of chronic cheilitis was retained.

The clinical evaluation demonstrated minimal edema and minor erythema of the lip, with crusting and peeling of white scale. Debridement of the scale demonstrated an otherwise normal lip surface (Fig. 1). Full body examination performed by dermatologists did not reveal any evidence of intraoral or cutaneous involvement. Her medical history was unremarkable. There was not a family history of psoriasis.

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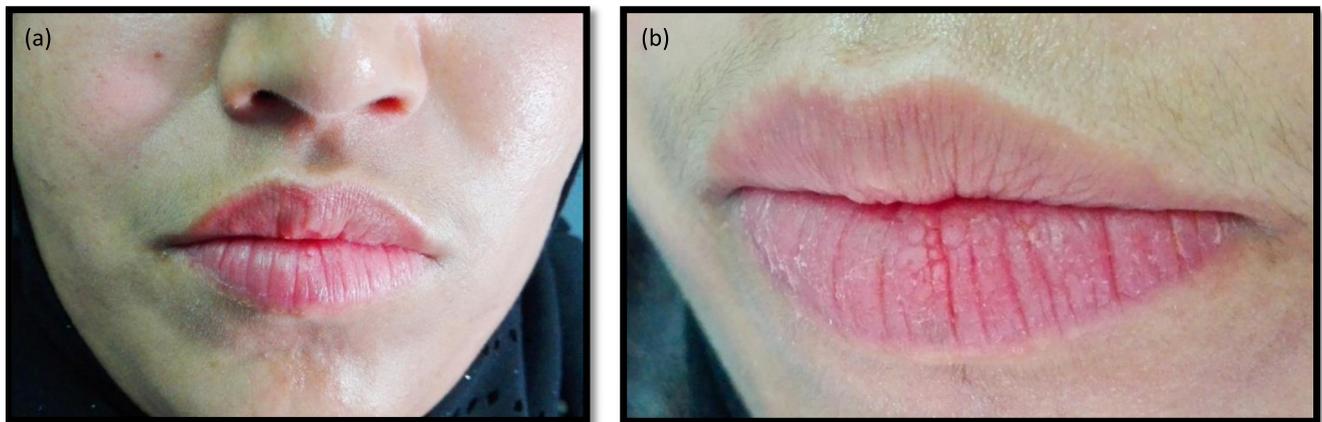


Fig. 1. (a) Mild edema and minimal erythema of both lips. (b) Cracking, fissure and adherent whitish scales on the vermillion.

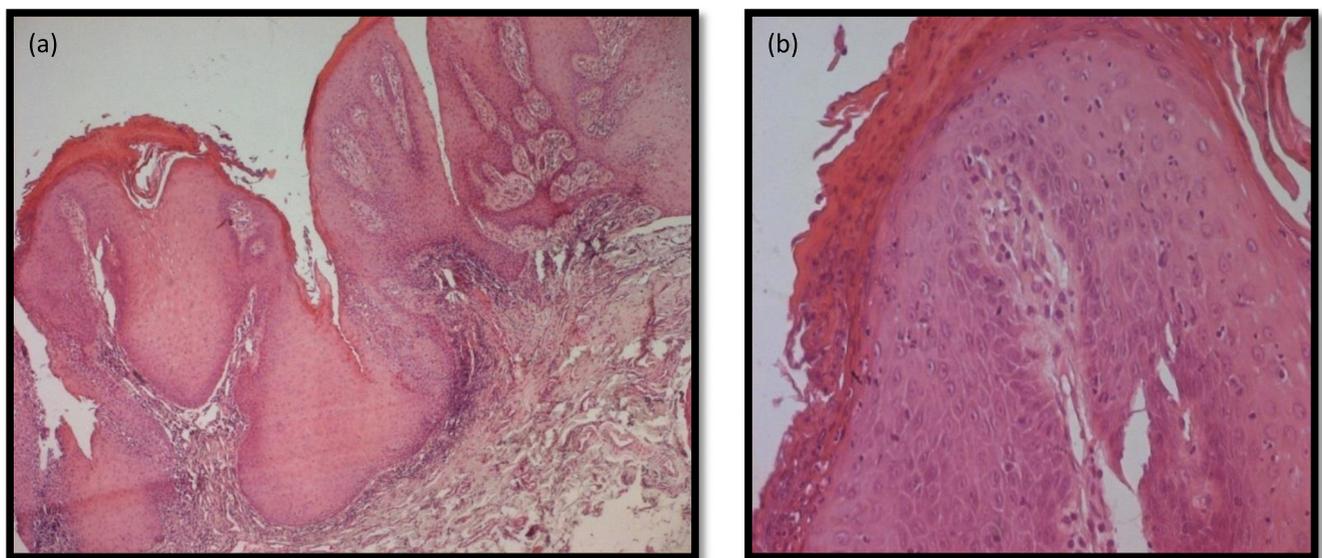


Fig. 2. (a) HE $\times 200$: Hyperplasia of the rete ridges and thinning of dermal papillae; Oedematous derm with dense cell infiltrate. (b) HE $\times 400$: Micro-abscesses of Munro (clusters of neutrophil polynuclear) within an hyperplastic and parakeratotic epithelial layer.

As these aesthetically disturbing lesions were chronic and persistent over the years, an obsession had overtaken our patient, resulting in a psychological setback. Her mother even said that she rarely came out of the house and she requested a labioplasty.

Many diagnoses were proposed such as contact cheilitis, actinic cheilitis, chronic eczema or actinic dermatitis. A biopsy was performed at the lower lip lesions for histological and direct immunofluorescence examination. Histologically, tissues showed a pattern consistent with psoriasis. Direct immunofluorescence was negative.

Tissues showed psoriasiform hyperplasia of the rete ridges and thinning of dermal papillae.

Acanthosis was observed. Oedematous derm was present with vasodilatation and dense cell infiltrate. We also had a collection of neutrophils in the upper epithelium within the parakeratotic epithelial layer (micro-abscesses of Munro). No granulomas were observed. Periodic acid-Schiff (PAS) stain for

fungal hyphae and direct immunofluorescence were negative (Fig. 2).

Our diagnosis concluded a case of oral psoriasis limited to the vermillion border of the lips with no identified family history or cutaneous involvement.

As a preliminary treatment, application of topic steroids (betamethasone dipropionate) twice a day for two weeks was prescribed. Additionally, we asked the patient to reduce physical manipulation of the lip. At the 15 days control, she reported a great decrease of pain and scales (Fig 3). Unfortunately, the patient was unreachable after this period.

Discussion

Psoriasis is a chronic inflammatory condition primarily affecting the skin. It is a frequently occurring disorder with a chronic and relapsing course. Approximately 1–3% of the global

Table I. Summary of cases of psoriatic cheilitis reported in the literature.

	Sex	Age	Skin lesions at presentation	Oral lesions	Treatment	Outcome	Exclusive lip involvement
Tosti <i>et al.</i> [9]	F	24	No	No	Steroid cream	CR	Yes
Rahman <i>et al.</i> [10]	F	20	No	No	Triamcinolone acetonide ointment	CR	Yes
Sehgal <i>et al.</i> [8]	F	16	No	No	Tacrolimus + calcipotriol + betamethasone dipropionate	CR	Yes
Ersoy-Evans <i>et al.</i> [11]	F	19	No	No	Fluticasone propionate 0.005% ointment	CR	No
Baz <i>et al.</i> [6]	F	22	No	No	Mometasone furoate 0.1%	CR	No
Apalla <i>et al.</i> [12]	M	20	No	No	Tacrolimus + salicylic acid	CR	Yes
	F	28	No	No	Tacrolimus + salicylic acid	CR	Yes
Blankinship <i>et al.</i> [1]	M	20	No	No	Tacrolimus + calcipotriol + betamethasone dipropionate	SI	Yes
Current case	F	21	No	No	betamethasone dipropionate	NA	Yes

Abbreviations: CR, complete response; F, female; M, male; SI, significant improvement; NA, not available.



Fig. 3. Appearance of the lips at 15 days of control showing a notable improvement.

population is affected with an equal gender distribution [2]. It is characterized by erythematous papules covered by silvery scales that gradually enlarge at the periphery, forming plaques.

Its etiology remains unknown, but it appears to be multifactorial with genetic and psychosomatic factors. Various triggers, such as trauma, infection and stress, may cause new episodes.

Common sites of involvement are the extremities, trunk, scalp and nails. The disease may occasionally involve genitalia and the anus. Although cutaneous lesions are generally sufficiently distinct to make a diagnosis, diagnostic difficulties can be found in cases of unusual involvement such as an oral location [2,3]. Indeed, oral psoriasis is a rare entity and remains a subject of controversy.

The existence of true psoriatic lesions of the oral mucosa is still discussed because neither the clinical nor the histological changes are absolutely specific. Literature reports only few

cases of psoriasis presenting exclusively oral lesions. Problems with the diagnosis of oral psoriasis arise from the fact that no distinction can be made on histological grounds between oral psoriasis and geographic tongue, geographic stomatitis, or the oral lesions of Reiter's syndrome (revealed by triad of symptoms: arthritis, nongonococcal urethritis and conjunctivitis).

Geographic tongue and geographic stomatitis may be an oral manifestation of psoriatic disease, presenting histopathological, immunohistochemical and genetic similarities with plaque psoriasis [4]. Unfortunately, their link with psoriasis remains unproven [2].

Despite these difficulties, there is some agreement among authors that oral psoriasis, although rare, does occur. Then, they propose strict criteria for the diagnosis: The clinical course of the oral lesions should parallel that of the skin lesions; an evocative histological examination (hyperplasia of the rete ridges, thinning of dermal papillae and micro abscesses of Munro) can be associated to a positive family history. Additionally, HLA typing has also been considered of great importance in supporting a diagnosis of oral psoriasis. The HLA antigens most frequently associated with psoriasis are B13, B17, B37, Cw4 and Cw6 [2,5].

Most cases of oral psoriasis reported in the literature have been associated with skin disease and/or intraoral involvement. Lesions may present on the buccal mucosa, tongue, gingiva, palate, and very rarely the lips and / or perioral area. To the best of our knowledge, there have only been six reported cases of isolated lip psoriasis (without intraoral or skin involvement) in the literature [6]. Table I summarizes the main epidemiological and clinical characteristics of all the cases reported to date, including ours.

Although rare, psoriasis of the lips can be the sole presentation of psoriasis, preceding the appearance of typical skin lesions by several years.

Certainly, diagnosis is best made when the clinical course of the oral lesions parallels that of skin lesions. However, in a long-standing eczema-like eruption occurring on the lips, psoriasis should be suspected in the differential diagnosis and a biopsy should be taken in case no response to treatment occurs [3].

In the present case, the histological examination showed features of psoriasis. There wasn't any chronic candidiasis and the clinical aspects ruled out Reiter's syndrome. Additionally, chronicity and clinical appearance of the lesions suggested also a psoriasis. Weighing against this diagnosis were the absence of mucosal or skin lesions, lack of a family history of psoriasis and the absence of HLA typing. Thus, in our case, diagnosis of isolated lip psoriasis was made only by the histopathological and clinical findings.

This case is therefore interesting, as it indicates that psoriasis can manifest with lip involvement only for a long time. Therefore, psoriasis should be considered in the differential diagnosis of chronic and/or recurrent, treatment-resistant oral mucosal lesions, even in the absence of absolute histopathological findings, accompanying skin lesions or a positive family history.

Mild trauma, chronic irritation or protruding teeth can lead to psoriatic lesions on the lips, especially in a genetically predisposed individual [3]. Brenner *et al.* [7] reported a case of psoriatic cheilitis triggered by protruding teeth, the condition was resolved by replacing the teeth with a non-irritating prosthesis. Lip psoriasis is worsened by cold or dry weather, physical manipulation such as lip biting and exuberant rubbing, and by irritating substances, suggesting the Koebner phenomenon. Most authors have reported a significant psychiatric morbidity and profound negative effects on emotional and social quality of life of their patients [1,3].

For this patient, although there were no cutaneous manifestations or other physical symptoms, her lip psoriasis was associated with substantial psychiatric morbidity.

Lip psoriasis can be clinically confused with contact cheilitis, chronic eczema, actinic dermatitis, chronic candidiasis and leucoplakia. In our case, these were not supported histologically.

Given the rarity of this condition, the prevalence, distribution, natural history, and most effective treatments are unknown. Although this condition is apparently unresponsive to mild topical steroids such as those frequently prescribed for the lips, it frequently responds well to more potent steroids. Additionally, the use of topical vitamin D analogues and tacrolimus has been beneficial [1]. Sehgal [8] advocates the use of a combined therapy based on topical tacrolimus, calcipotriol, and betamethasone dipropionate.

Conclusion

Perioral psoriasis is an unusual presentation of psoriasis. Our case demonstrates the significant diagnosis difficulties of psoriasis based upon exclusive lip lesions. Then it is suggested that psoriasis should be in the differential diagnosis of lip lesions presenting with fissuring and scale, especially in the setting of a personal or family history of psoriasis, although these are not always present. Clinical suspicion is required to detect this disease because it may be confused with more common conditions, such as candidiasis, irritant or actinic cheilitis, bacterial infection or atopic dermatitis.

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

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