Looking Beyond the Cyclosporine “Swish and Spit” Technique in a Recalcitrant Case of Erosive Lichen Planus Involving the Tongue

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Abstract
Oral lichen planus is a relatively common autoimmune disease affecting the middle-aged population. Although no treatment is necessary for a benign asymptomatic case, in case of erosive lichen planus, topical corticosteroids form the mainstay of treatment. In case of failure, apart from tacrolimus, cyclosporine, using the “swish and spit” technique, is a valid therapeutic intervention. In our case, though, this therapeutic option had to be replaced by the systemic use of cyclosporine with gratifying results.
Introduction

Lichen planus (LP) is a chronic mucocutaneous disorder of the stratified squamous epithelium affecting oral and genital mucous membranes, the skin, nails, and scalp. LP is estimated to affect 0.5–2.0% of the general population, with a prevalence as high as 2.6% in the Indian population. Oral LP (OLP) is comparatively more frequent than the cutaneous form, and it is more persistent and resistant to treatment and afflicts the middle-aged population, especially tobacco chewers and smokers [1]. The buccal mucosa, tongue, and gingiva are commonly affected sites in OLP [2]. The clinical presentation of OLP classically consists of 6 types: reticular, atrophic, papular, bullous, plaque, and erosive or ulcerative type [3]. Although it is often asymptomatic, the erythematous and erosive forms can cause severe pain, interfering with eating and swallowing and thus requiring therapy.

Here, we present a case report of a male patient with debilitating tongue LP which was resistant to various topical and systemic treatments and was successfully treated with cyclosporine oral solution.

Case Report

A 32-year-old male patient presented with a burning sensation in the mouth for 12 months. The complaint was insidious in onset and aggravated on taking spicy food. The personal history revealed that the patient was an occasional drinker and smoker and was suffering from occupation-related stress. The medical history and family histories were unremarkable.

Physical examination revealed a large erythematous plaque on the dorsum of the tongue (Fig. 1) with violaceous lacelike lesions on the buccal mucosa and no affliction of the skin, other mucosa, and nails. A provisional diagnosis of OLP was made based on the history and clinical findings. The patient revealed that he had been treated earlier with topical betamethasone 0.05%, tacrolimus 0.1%, tablet dapsone 100 mg for 6 weeks, and tablet methotrexate 12.5 mg once a week for 4 weeks with no appreciable response.

Histopathological examination of the lesion was performed to confirm the diagnosis and to rule out any malignant transformation which was consistent with LP. Laboratory screen for hepatitis B and C and baseline hemogram, liver function tests, and kidney function tests were within normal limits. The patient was started on cyclosporine (Psorid Biocon™) oral solution 100 mg/mL as a “swish and spit” medication, 3 times daily, each treatment lasting for 5 min. However, there was no significant improvement in the lesions after 5 weeks of therapy, and the patient was then asked to take cyclosporine 200 mg/day orally (1 mL of solution b.d.) mixed with water, milk, or juice (except grape fruit juice) 1 h before meals. The decision on giving the solution in preference to the capsule was based on the cost effectiveness of the solution and the fact that accurate dosing according to the bodyweight can be achieved by the oral solution. After 6 weeks of treatment with systemic cyclosporine, there was a dramatic response (Fig. 2) with near complete resolution of the lesion. The patient
was asked to continue the therapy at a dose of 1 mL a day for another month, which resulted in complete resolution of the lesion (Fig. 3). The patient reported no adverse event connected with the use of cyclosporine.

**Discussion**

Treatment of OLP is aimed primarily at reducing the length and severity of symptomatic outbreaks, while a more pressing need is to prevent malignant transformation of OLP. No medication is necessary for the benign form of OLP; however, high-potency topical corticosteroids are the mainstay of treatment in cases presenting with severe pain and/or a burning sensation amongst other options (Table 1).

The pathogenesis of OLP is based on the interplay of helper/inducer T cells and the antigen-presenting cells, leading to the activation of auto-cytotoxic CD8+ T cells that release interferon-γ which triggers the apoptosis of the basal cells of the oral epithelium [4, 5]. Cyclosporine has been used via its action on the helper/inducer T lymphocyte and inhibition of the CD1+ and CD14+ antigen-presenting cells [6]. Moreover, the drug modulates cytokines, such as IL-2 and interferon-γ, leading to a reduced function of effector T cells that play a role in OLP. The failure of other systemic agents (dapsone and methotrexate) made cyclosporine a logical choice in our case [7].

For treating OLP, cyclosporine has been used as a topical therapy in the past, using the “swish and spit” technique. However, the results of the latter technique, though seemingly safe, are variable. This is because cyclosporine is inactivated by cytochrome P450-dependent biotransformation in the mucosa [8, 9]. Also, as the purported mode of action is by its action on systemic T lymphocytes, it is unlikely that the "swish and spit" technique can affect a major fraction of the T-cell population, since optimal systemic levels are not attained by topical cyclosporine [10]. This could explain the initial failure of cyclosporine in our patient.

Of the few reports of the use of oral cyclosporine in the treatment of OLP, a similar case was reported by Boyce et al. [11] who used systemic cyclosporine in a dose of 150 mg b.d. for severe recalcitrant erosive mucosal LP and found a marked improvement in genital and oral erosion/ulceration within 6 weeks. No adverse event was reported with cyclosporine by these authors, as was seen in our case.

Our case highlights that though systemic cyclosporine is an effective third-line modality for erosive LP, probably a systemic use is better than the “swish and spit” technique.

**Statement of Ethics**

The authors have no ethical conflicts to disclose.

**Disclosure Statement**

There are no sources of support or conflicts of interest.
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Fig. 1. A large erythematous plaque present on the dorsum of the tongue at the first visit.
Fig. 2. Improvement of the tongue lesion after initiation of oral cyclosporine solution (200 mg/day).
**Fig. 3.** Complete resolution of the lesion after 3 months of treatment with systemic cyclosporine.

**Table 1.** Treatments used for erosive lichen planus with tongue involvement

|   |                                                                 |
|---|-----------------------------------------------------------------|
| 1 | Topical and systemic corticosteroids [12]                       |
| 2 | Tacrolimus and pimecrolimus [13]                                |
| 3 | Topical and systemic retinoids [14]                             |
| 4 | Oral and topical cyclosporine A [15]                            |
| 5 | Dapsone [16]                                                    |
| 6 | CO₂ laser [17]                                                  |
| 7 | Photodynamic therapy [18]                                      |
| 8 | Cryosurgery [19]                                                |
| 9 | Ozone therapy [20]                                              |