Topical tacrolimus for refractory superficial mucoceles in a patient with chronic graft versus host disease

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INTRODUCTION

The common mucocele is a painless, bluish nodule that forms from subepithelial mucosal extravasation after damage to a minor salivary gland duct.[1] These lesions typically occur on the lips secondary to biting trauma and resolve spontaneously.[2] Although morphologically similar, superficial mucoceles are clinically and pathologically distinct. Superficial mucoceles present as multiple recurrent, fragile, translucent vesicles in the oral cavity resembling herpes simplex or bullous lichen planus with characteristic location of disease on the soft and hard palate, retromolar region, and buccal mucosa.[1] While pathogenesis is unknown, an inflammatory basis is presumed because of its association with lichen planus and chronic graft-versus-host disease (cGVHD).[1,2] Only a handful of cases of superficial mucoceles have been reported, and most are asymptomatic and with a self-limiting disease course. Although symptomatic cases are rare, they are problematic because of lack of safe, simple, and efficacious treatment options. Importantly, such cases typically occur in cancer patients; thus, treatment is essential to maintaining quality of life in patients with multiple concurrent symptomatic comorbidities. We present a case of refractory painful superficial mucoceles in a patient with cGVHD successfully treated with topical tacrolimus swish-and-spit.

Abbreviation used:

cGVHD: chronic graft-versus-host disease

CASE REPORT

The patient is a 72-year-old man with a history of peripheral T-cell lymphoma post radiotherapy, multiagent chemotherapy, and haploidentical allogeneic stem cell transplant on December 1, 2017. He had biopsy-proven cGVHD of the skin and oral mucosa diagnosed in June 2018 after presenting with pink papules on the trunk and extremities as well as lichenoid changes on the buccal and labial mucosa. His orocutaneous disease was managed successfully with a tapered course of oral corticosteroids. Later that month the patient was also found to have herpes simplex virus esophagitis treated with acyclovir, and repeat biopsies and culture were negative after treatment. On follow-up 3 months later, the patient complained of persistent oral pain and dysphagia unresolved with topical dexamethasone. On examination, the patient had soft palate and pharyngeal erythema with numerous clear vesicles and round erosions as well as lichenoid patches on the buccal mucosa (Fig 1). Superficial mucoceles were favored, yet herpes simplex virus polymerase chain reaction was rechecked given the vesicular appearance and the patient's history; the results were negative. Because of the exuberance of disease and unresponsiveness to oral steroids, the dental GVHD specialist at the National Institutes of Health was consulted who recommended a stronger formulation of oral steroids. Unfortunately, insurance coverage was denied, and, subsequently, tacrolimus swish-and-spit was prescribed. The patient dissolved 1 mg of tacrolimus in 1 L of water using this solution to swish-and-spit twice per day. The patient

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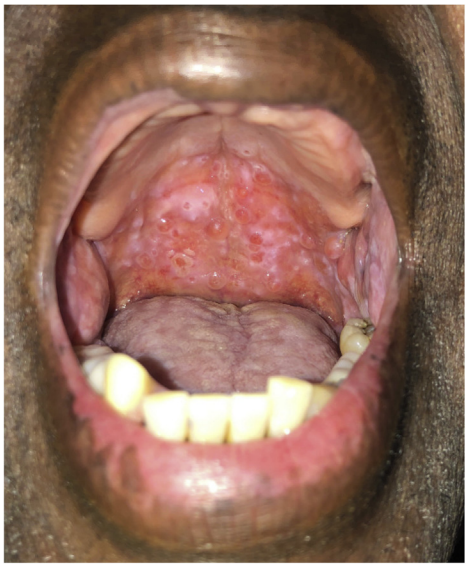

Fig 1. Multiple superficial mucoceles. Clinical presentation on initial dermatologic consultation with multiple clear vesicles and round erosions on an erythematous soft palate and pharynx as well as lichenoid patches on the buccal mucosa.

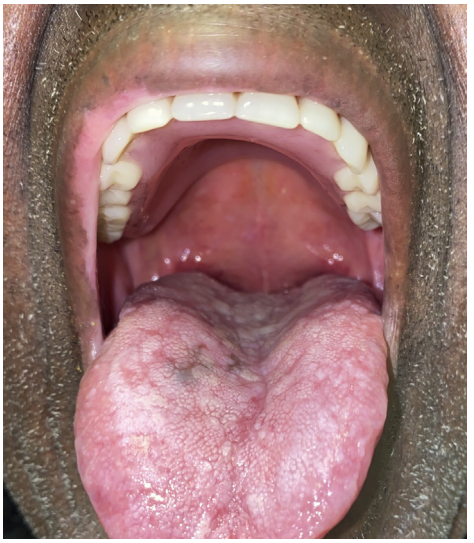

Fig 2. Resolution of superficial mucoceles. Clinical appearance after 3 months of topical tacrolimus swish-and-spit treatments.

experienced progressive improvement of lesions with complete resolution after 3 months of treatment (Fig 2).

DISCUSSION

Tacrolimus is a calcineurin inhibitor that can be used topically for the treatment of inflammatory and immune-mediated oral lesions including lichen planus, benign migratory glossitis, and cGVHD.[3,4] Swish-and-spit treatments are low cost and easy to perform, which makes topical tacrolimus an excellent nonsystemic treatment option for mucosal inflammatory disease. As such, its application in the treatment of superficial mucoceles is sensible. Previously, superficial mucoceles have been treated with laser vaporization, intralesional or topical corticosteroids, cryotherapy, and γ-linoleic acid.[5] Aside from corticosteroids, none of these treatments addresses the inflammatory nature of disease, relying on generalized destructive mechanisms instead. Further, although corticosteroids are reliable in isolated disease, mucoceles associated with cGVHD have been historically unresponsive to this treatment.[1,5] This resulting lack of therapeutic options, specifically in patients with commonly recurrent and refractory disease, is problematic, yet the option of topical tacrolimus provides a reasonable and efficacious solution that should be considered in such cases.

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