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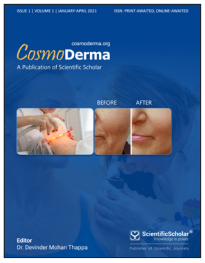

Letter to the Editor

Primary complex aphthosis responding well to dapsone

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Dear Sir,

A severe type of recurrent aphthous stomatitis known as complex aphthosis is characterized by the presence of three or more oral ulcers almost always, either with or without genital aphthosis. Due to its severity and intractability, the treatment is sometimes difficult and frequently necessitates the use of systemic medicines. We present a case of complex aphthosis with an excellent response to dapsone.

A 36-year-old male patient presented with complaints of recurrent multiple painful oral ulcers that had been bothering him for the past 9 months, as well as difficulty opening his mouth, swallowing, and eating. The patient had 5–6 episodes of oral ulceration that healed on their own and with treatment with topical medicines. He also had multiple painful genital ulcers on the glans for the past 4 months. He has been married for 8 years, is heterosexual, and has no similar complaints in his partner. On oral examination, there were multiple ulcers with well-defined margins, ranging from 1 mm to 10 mm, with a peripheral erythematous halo and yellowish to greyish floor on the labial mucosa, under surface of the tongue, gingiva, and hard palate [Figure 1]. On genital examination, there were multiple round to ovoid ulcers of diameter 2–4 mm with punched-out edges, a yellowish to grayish floor, yellowish crusting at places, non-indurated, and tender on touch on glans penis with no inguinal lymphadenopathy [Figure 2]. There was no history of fever, weakness, fluid-filled lesions, drug use, gastrointestinal, neurological, ocular, or joint pain. The Grams stained smear revealed only a few pus cells. Tzanck smear, KOH studies, dark ground microscopy, pus culture and sensitivity, Herpes simplex virus polymerase chain reaction, HIV serology, and VDRL were negative. Pathergy testing, HLA-B51, serum iron, folate, and vitamin B12 levels came out as non contributory. Histopathology showed a mixed infiltrate consisting mostly of neutrophils along with lymphocytes and macrophages at the base of the ulcer. Features of small vessel vasculitis were also seen. A diagnosis of primary idiopathic complex aphthosis after the exclusion of Behcet's disease was made. The patient was started on dapsone 100 mg twice daily after G6PD screening along with topical triamcinolone gel. The lesions responded well, and all lesions healed in 1 month.

Jorizzo *et al.* coined the term "complex aphthosis" in 1984 to describe recurrent oral and genital aphthous ulcers or almost constant three or more oral aphthae without other manifestations of Behcet's disease.[1] Later, it might turn into Bechet's disease, as mucocutaneous ulcers are frequently the first signs of Bechet's disease. Complex aphthosis is a poorly understood condition with a hazy etiopathogenesis. Genetics, trauma, smoking, mental stress, hormonal factors, viruses, bacteria, food sensitivity, and immunological dysregulation are just a few of the possible causes.[2] Complex aphthosis can manifest as either a idiopathic(primary) or secondary to systemic disease. The presentation is of small, painful, shallow, and round-to-oval ulcers that are surrounded by a yellowish pseudomembrane with a sharp, red border on the oral mucosa, and genitalia. For the

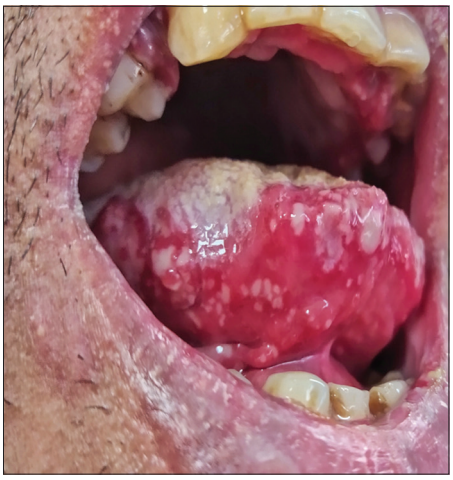

Figure 1: Multiple ulcers with well-defined margins, ranging from 1 mm to 10 mm, with a peripheral erythematous halo and yellowish to greyish floor on the labial mucosa, undersurface of the tongue, gingiva, and hard palate.

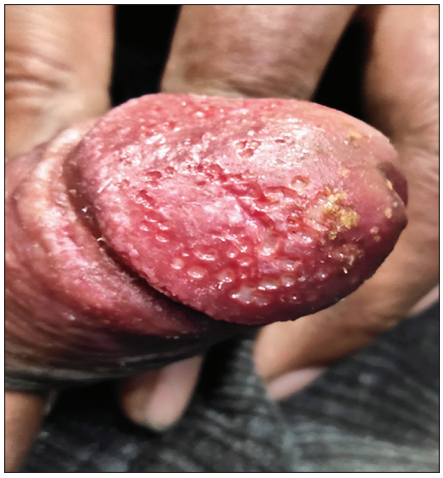

Figure 2: Multiple round to ovoid ulcers of diameter 2–4 mm with punched-out edges, a yellowish to grayish floor, and yellowish crusting at places on glans penis.

diagnosis and management of complex aphthosis, there is no established algorithm. The treatment options vary; however, systemic medications should generally be used for most patients. An effective systemic medication that is frequently used is oral prednisone.[3] Other drugs that are studied are colchicine, dapsone, pentoxifylline, methotrexate, thalidomide, and montelukast.[4,5] Due to the chronic nature of the disease, dapsone and colchicine are better options for treatment than steroids and immunosuppressants. Neutrophilic chemotaxis and activity are known to be important in pathogenesis. Furthermore, the presence of neutrophils in the biopsy of our patient prompted us to start him on dapsone. Dapsone is a relatively safe medication that can be used and has an antineutrophilic effect. Without any negative side effects, our patient tolerated dapsone well.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Conflicts of interest

There are no conflicts of interest.

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