

Melkersson-Rosenthal Syndrome: Unusual Extraintestinal Manifestation of Crohn's Disease

Patrícia Araújo^{1*}, Ana Frederica Parente¹ and Alda Andrade²

¹Internal Medicine Department, Unidade Local de Saúde do Alto Minho, Portugal

²Gastroenterology Department, Unidade Local de Saúde do Alto Minho, Portugal

***Corresponding author:** Patrícia Araújo, Internal Medicine Department, Unidade Local de Saúde do Alto Minho, Portugal.

E-mail: araajo.patricia0102@gmail.com

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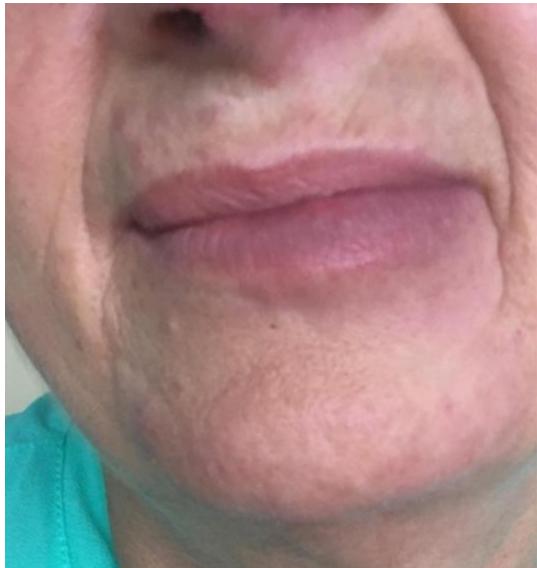


Figure 1 and 2: Asymmetrical edema on the left lower lip.

Clinical Image

A 62-year-old woman with Crohn's disease diagnosed in 2013, receiving azathioprine and infliximab, developed right-sided peripheral facial paralysis and left lower lip edema in 2018 (Figure 1 and 2). In 2019, she was referred to an internal medicine outpatient clinic for further evaluation and was suspected of having Melkersson-Rosenthal syndrome, a condition sometimes associated with inflammatory bowel disease. The diagnosis was confirmed by biopsy of the lip lesion.

Treatment was initiated with oral prednisolone at a dose of 60mg, but successive relapses occurred during dose tapering. Dapsone therapy was subsequently attempted; however, there was no significant improvement in the lip lesion, and the patient developed hemolytic anemia, leading to discontinuation of the drug. Given the ongoing immunosuppressive therapy, refractoriness to treatment, and adverse effects of systemic corticosteroids, a literature review was performed. Intralesional triamcinolone administration was then selected, resulting in complete resolution of the edema.

Melkersson-Rosenthal syndrome is a rare disorder of uncertain etiology, characterized by a triad of recurrent orofacial edema, intermittent facial paralysis, and tongue abnormalities [1,2]. Although oral involvement is common in Crohn's disease, lack of response to standard treatment and presence of additional symptoms should prompt consideration of alternative diagnoses, as illustrated in this case, allowing for subsequent effective targeted therapy.

Keywords: Melkersson-rosenthal syndrome; Crohn disease; Angioedema

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