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Orofacial Swelling and Elevated Fecal Calprotectin Levels in a Patient with Melkersson-Rosenthal Syndrome

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Background: Melkersson-Rosenthal syndrome (MRS) is a rare, inherited neurocutaneous syndrome characterised by a triad of clinical features: recurrent facial nerve palsy, persistent or recurrent orofacial swelling, fissured tongue. These three clinical entities can be present all together or not (complete or incomplete forms).

Sometimes MRS may be associated with Crohn's disease or sarcoidosis.

Case History: A 25-year-old male presented with recurrent acute abdominal pain, persistent upper lip edema and a history of allergic rhinitis and bronchial asthma. Intraoral examination revealed a fissured tongue. Oral mucosa biopsy demonstrated non-caseating granulomas. Fecal calprotectin test depicted elevated levels. Complete abdominal ultrasound and Magnetic Resonance Enterography (MRE) were performed but did not highlight anything significant. Other systemic granulomatous diseases were excluded.

The patient was treated with oral corticosteroid with partial and transient benefit. Then intralesional treatment with triamcinolone was administered, with good results.

The patient was advised to repeat calprotectin dosage after some months, in order to exclude or confirm the hypothesis of a secondary form of facial granulomatosis.

Discussion: MRS is a rare neurocutaneous disease. MRS diagnosis and treatment are difficult due to its rarity and unclear etiology.

Early recognition of MRS can reduce diagnostic delays, which often lead to suboptimal patient outcomes, and facilitate appropriate management.

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