

# Melkersson–Rosenthal Syndrome: The Role of The Dentist In Early Diagnosis: A Case Report

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## Abstract

Melkersson–Rosenthal syndrome (MRS) is a rare disorder that belongs to the group of orofacial granulomatosis. It is classically defined by a clinical triad consisting of recurrent orofacial edema, peripheral facial nerve palsy, and fissured tongue, although the complete form is uncommon. Diagnosis relies mainly on histopathological examination, which demonstrates non-caseating granulomatous inflammation. The etiology and pathophysiology of MRS remain unclear, and current treatments are essentially symptomatic, with variable efficacy and frequent recurrences. Dentists play a crucial role in early diagnosis, as oral manifestations are often the first signs of the disease. This article reports a clinical case and discusses the diagnostic approach and therapeutic management of MRS based on a review of the literature.

**Keywords:** Facial Palsy; Histopathology; Macrocheilitis; Melkersson–Rosenthal Syndrome; Orofacial Granulomatosis.

## 1. Introduction

Melkersson–Rosenthal syndrome (MRS) is a rare neuro-muco-cutaneous disorder characterized by the association of recurrent orofacial edema, peripheral facial nerve palsy, and fissured tongue, although the complete triad is infrequently observed, making diagnosis challenging [1].

The etiology of MRS remains unclear, with immuno-allergic, infectious, and genetic mechanisms being the main proposed hypotheses [2 - 4]. Histopathological findings of non-caseating granulomatous inflammation are central to supporting the diagnosis and excluding other granulomatous conditions [3].

This article reports a rare case of Melkersson–Rosenthal syndrome presenting with the complete clinical triad and is accompanied by an updated review of the literature, with particular emphasis on the role of the dentist in early diagnosis.

## 2. Case Report

A 24-year-old female patient was referred to the Department of Oral Pathology and Surgery for a labial biopsy due to persistent lip swelling of unknown etiology. The patient reported a five-year history of chronic labial edema predominantly affecting the upper lip, characterized by recurrent paroxysmal episodes without any identified triggering factors (Fig. 1). These episodes were not associated with laryngeal edema or systemic allergic manifestations.



**Fig. 1:** Clinical Appearance of Labial Edema. Diffuse, painless swelling of the upper and lower lips involving both the vermilion and cutaneous portions, without skin discoloration.

Her medical history revealed a unilateral peripheral facial palsy that had occurred five years earlier, followed by two recurrences. Clinical examination showed persistent facial paresis (Fig. 2), associated with ipsilateral epiphora and hyperhidrosis. The patient reported no gastrointestinal symptoms suggestive of Crohn's disease and had no history of tuberculosis. Extraoral examination revealed a painless, elastic edema involving both the upper and lower lip (Fig. 1), affecting the vermilion and cutaneous lip without discoloration. Facial asymmetry was evident during facial movements, particularly while blowing (Fig. 2), with right-sided facial paresis. No dysphagia or cervical lymphadenopathy was observed.



**Fig. 2:** Peripheral Facial Nerve Paresis. Facial asymmetry observed during facial movement (blowing maneuver), consistent with right-sided peripheral facial nerve paresis.

Intraoral examination showed obliteration of the anterior maxillary vestibule and localized gingival inflammation in the maxillary incisor region, associated with dental plaque accumulation. The tongue presented a fissured appearance with characteristic transverse grooves, consistent with a plicated tongue (Fig. 3).

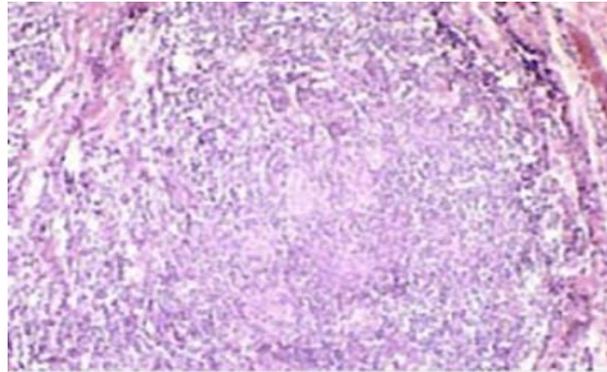


**Fig. 3:** Fissured Tongue. Enlarged tongue showing multiple transverse fissures.

The patient was referred for an otorhinolaryngology consultation to investigate the etiology of the facial palsy. Given the association of labial edema, fissured tongue, and a history of facial paralysis, Melkersson–Rosenthal syndrome was suspected. A labial biopsy (Fig. 4) was performed and confirmed the diagnosis, revealing multiple non-caseating epithelioid granulomas with multinucleated giant cells, associated with a perigranulomatous lymphoplasmacytic infiltrate (Fig. 5).



**Fig. 4:** Labial Biopsy Procedure. A biopsy specimen was obtained from the lower lip for histopathological examination.



**Fig. 5:** Histopathological Findings (Hematoxylin–Eosin Staining,  $\times 10$ ). Non-caseating epithelioid and multinucleated giant cell granulomas located in the superficial connective tissue, surrounded by a lymphocytic infiltrate, consistent with granulomatous cheilitis.

Additional investigations were conducted to exclude systemic causes, including tuberculin skin testing, chest radiography, complete blood count, erythrocyte sedimentation rate, and assessment of iron, folic acid, and vitamin B12 levels. No biological inflammatory syndrome or signs of malabsorption were identified.

An immunological and endocrine evaluation performed by an internist showed normal angiotensin-converting enzyme levels, negative antinuclear antibodies and ANCA, normal complement fractions, and normal total IgE levels. Mild hypothyroidism was detected, without associated antithyroid antibodies.

Based on the presence of the complete clinical triad and the exclusion of other etiologies, the diagnosis of Melkersson–Rosenthal syndrome was established. Systemic corticosteroid therapy with prednisone at a dose of 30 mg/day was initiated for one month.

Clinical follow-up was carried out over a period of 12 months. A marked regression of labial edema and significant improvement of facial nerve function were observed within the first weeks of treatment. After gradual tapering and discontinuation of corticosteroid therapy, the patient experienced intermittent mild recurrences of labial swelling, which were less severe and shorter in duration than previous episodes and did not require reintroduction of systemic corticosteroids.

At the last follow-up visit, facial paralysis had partially resolved, with no progression of neurological symptoms. The patient remained under regular multidisciplinary surveillance, including dental and otorhinolaryngological follow-up, with reinforcement of oral hygiene measures and elimination of local inflammatory factors.

### 3. Discussion

Melkersson–Rosenthal syndrome (MRS) is a rare granulomatous disorder classified within the spectrum of orofacial granulomatoses. Its low prevalence and the frequent absence of the complete clinical triad contribute to delayed diagnosis and underrecognition in clinical practice [1], [4]. The present case illustrates these diagnostic challenges, as the patient experienced several years of isolated and sequential manifestations before the syndrome was recognized.

The clinical presentation of MRS is highly heterogeneous. Although the classical triad consists of recurrent orofacial edema, peripheral facial nerve palsy, and fissured tongue, all three features are simultaneously present in only a minority of patients [4 - 6]. Orofacial edema is the most constant and usually the earliest manifestation, often leading patients to consult dental rather than medical services. Chronic or recurrent edema may progress to persistent macrocheilitis with functional and aesthetic consequences, as observed in the present case [6 - 11].

Intraoral findings, including fissured tongue, gingival inflammation, and vestibular obliteration, may provide important diagnostic clues. While a fissured tongue is frequently asymptomatic and nonspecific, its association with recurrent labial edema should prompt consideration of MRS [4], [10]. These features highlight the crucial role of dental practitioners in early recognition and referral.

Peripheral facial nerve palsy occurs in approximately one-third of MRS cases and may be recurrent or persistent [12]. Proposed mechanisms include inflammatory compression of the facial nerve within the facial canal and granulomatous infiltration of nerve fibers, reflecting the neuro-inflammatory nature of the disease [13], [14]. In the present case, facial nerve involvement preceded the diagnosis by several years, illustrating the temporal dissociation commonly reported in the literature.

The etiopathogenesis of MRS remains incompletely understood. Genetic susceptibility, immune dysregulation, allergic mechanisms, and infectious triggers have all been suggested [2], [4]. Chronic antigenic stimulation has been proposed as a potential mechanism leading to granuloma formation in orofacial tissues [7]. The possible contribution of odontogenic and periodontal inflammatory foci has been discussed, as their elimination may reduce local inflammatory burden, although definitive causal relationships have not been established [6].

Histopathological examination remains a key element in confirming the diagnosis. Typical findings include non-caseating epithelioid granulomas with multinucleated giant cells and a perigranulomatous lymphoplasmacytic infiltrate [3], [8]. However, granulomatous features may be absent in early stages, emphasizing the need for clinicopathological correlation and, in some cases, repeat biopsy. A wide range of conditions must be

excluded, including Crohn's disease, sarcoidosis, infectious granulomatous diseases, allergic macrocheilitis, vascular malformations, and Miescher's granulomatous cheilitis [15–17].

Therapeutic management of MRS remains challenging due to the absence of standardized treatment protocols. Systemic corticosteroids are considered first-line therapy and may lead to significant improvement in orofacial edema and facial nerve dysfunction [4], [5]. In the present case, corticosteroid therapy resulted in marked clinical improvement; however, mild recurrences were observed during follow-up, consistent with the relapsing course commonly described in the literature. Intralesional corticosteroids and immunomodulatory agents have been proposed for refractory or localized forms, with variable outcomes [15]. Surgical interventions are reserved for severe, persistent, or disfiguring cases and should be approached cautiously due to the risk of recurrence [2], [14].

This case underscores the importance of long-term follow-up and multidisciplinary management. Dentists occupy a central position in the diagnostic pathway, as oral and perioral manifestations are often the earliest signs of MRS. Early suspicion, appropriate biopsy, elimination of odontogenic inflammatory factors, and close collaboration with otorhinolaryngologists, dermatologists, neurologists, and internists are essential to optimize patient outcomes and limit disease progression.

#### 4. Conclusion

Melkersson–Rosenthal syndrome is a rare and frequently underdiagnosed disorder owing to its variable and often incomplete clinical presentation. Diagnosis relies on careful clinical assessment supported by histopathological findings. Given that oral and perioral manifestations are commonly the earliest signs, dentists play a key role in early recognition and in initiating appropriate multidisciplinary management. A better understanding of the underlying pathophysiological mechanisms is needed to support the development of more targeted and effective therapeutic approaches and to improve long-term patient outcomes.

#### 5. Conflict of Interest

The authors declare that they have no conflict of interest.

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