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# Recurrent Facial Palsy in an Adolescent - Case Report

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## ABSTRACT

Recurrent facial nerve paralysis is an unusual occurrence.

Recurrent facial palsy, oro-facial swelling, and a fissured tongue. Lower Motor Neuron Palsy. No definitive therapy is available, and frequent recurrences are common.

Idiopathic recurrent Lower Motor Neuron facial palsy with atypical presentations can be indicative of Melkerson-Rosenthal syndrome.

## Introduction

Paralysis of the facial nerve is a non-progressive neurological disorder, and its recurrence is unusual [1]. The causes predisposing to recurrent idiopathic Lower Motor Neuron (LMN) facial palsy are not well understood [1]. Apart from viral and immune-mediated pathogenesis, a genetic predisposition has also been proposed [1]. A history of recurrent LMN facial palsy, orofacial swelling, and/or a fissured tongue could indicate Melkerson-Rosenthal syndrome (MRS) [2]. MRS is a rare, neurocutaneous disease of unknown etiology. The incidence reported is between 0.2 and 80 in 100,000 per year [3]. The classical triad can be seen in only 8% to 18% of patients. A review of literature has shown that this syndrome can manifest in a number of ways, usually not fulfilling the criteria of the triad, which is why it is often ignored and misdiagnosed [4, 5]. In this case report, a patient with MRS who had complaints related to recurring facial palsy is presented.

## Patient Information

A 15-year-old male presented to the clinic with a history of acute-onset weakness affecting the left half of his face for one month. This was associated with deviation of the mouth to the opposite side, incomplete closure of the left eye, difficulty in eating, and drooling on the left side. There was no history of trauma, fever, rash, earache, vertigo, hyperacusis, or exposure to cold stress prior to the episode. There was no history of allergy or atopy in the patient or

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his family. A past history of two similar episodes on the same side in the last 5 years was present, which had resolved completely over 4-5 weeks with oral medications.

## Clinical Findings

Upon examination, the patient had left-sided LMN Grade-4 House Brackman facial palsy. The patient also had a deeply furrowed tongue and mild edema and swelling over the left upper lip and buccal mucosa. The edema was non-erythematous, painless, and non-pruritic. The otolaryngological and systemic examinations were normal. The neurological examination, including other cranial nerves, was normal. Based on the history of recurring facial palsy, folds and furrows in the tongue, and swelling of the face and lips, a clinical diagnosis of MRS was considered. A family history of a fissured tongue in the other siblings added to the suspicion of MRS.

## Timeline

The patient had experienced two similar episodes on the same side in the last five years. Both of these episodes were treated with oral medications for a duration of approximately 4 to 5 weeks each.

## Diagnostic Assessment

### On PE, the patient had Left sided LMN facial palsy with fissured tongue and oro-facial edema

The patient's complete blood count, blood sugar, blood urea, and other biochemical investigations were all normal. His serum ACE levels were also within the normal range. A high-resolution computed tomography (HRCT) of the temporal bone showed no abnormalities.

### Therapeutic intervention

The patient was started on an oral steroid, Prednisolone, in tapering doses. The regimen began at 1mg/kg/day, administered 12 hourly for 2 weeks. This was followed by a reduced dosage of 0.5mg/kg/day over the next 2 weeks, with a plan to further taper and stop in the subsequent 2 weeks. In addition to this, eye care and physiotherapy were advised.

### Follow up and Outcome

At the 4-week follow-up, the oro-facial swelling had subsided and a partial improvement in weakness

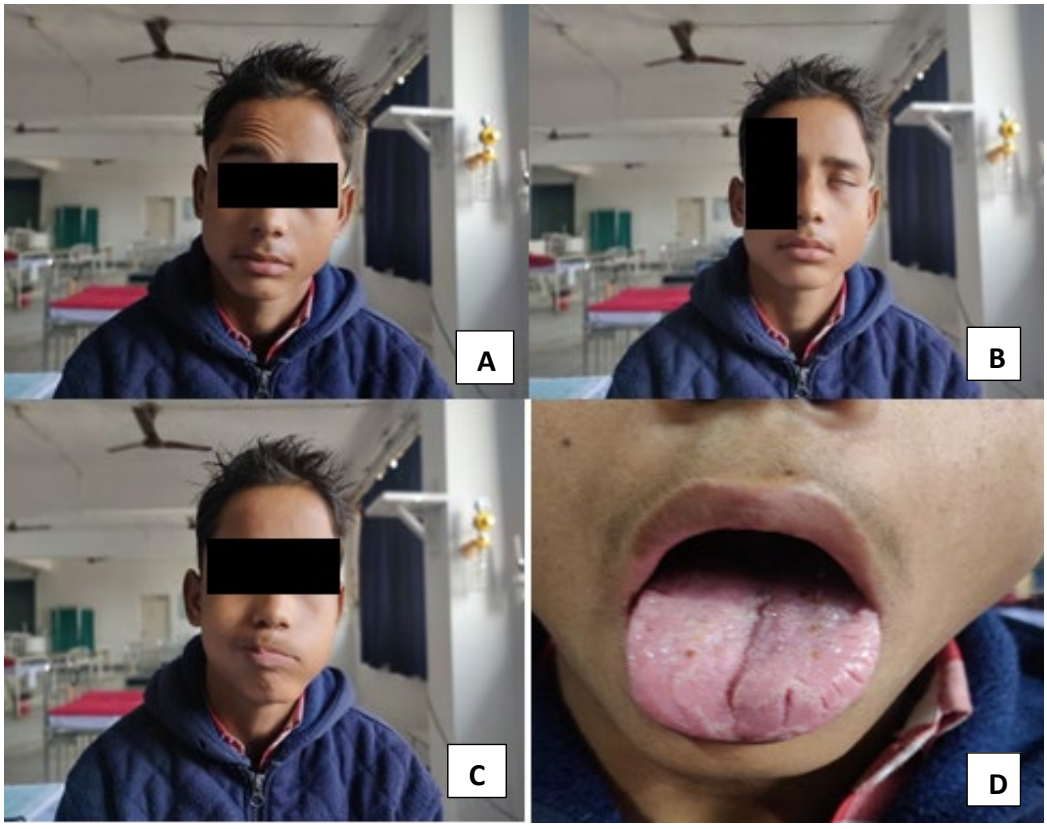


Fig. 1: (A-C): Patient showing features of lower motor neuron palsy (D): Patient with fissured tongue

was observed. The patient's compliance with the treatment was good, and there were no effects of steroid toxicity observed, except for weight gain and increased appetite.

## Discussion

Melkerson-Rosenthal was first described in 1928 as peripheral facial paralysis and swelling of the lips. In 1931, Rosenthal completed the triad by adding the presence of a fissured tongue [6]. The age at onset varies from early to late childhood. A monosymptomatic or oligosymptomatic form (presence of 2 symptoms) is more common [1]. Although its etiology is unknown, a familial tendency of incomplete autosomal dominance, chromosomal mutation, and fibrous dysplasia of the temporal bone have been suggested as the etiologic factors for recurrent facial palsy [1, 5]. Various factors like infections, immune deficiency, food intolerance, and stress may contribute [7]. A genetic predisposition is suspected because several cases are reported within the same family [5]. Furthermore, lingua plicata, Bell's palsy, and angioedema each can be familial [5]. MRS resembles angioedema but it can be differentiated by the lack of response to anti-histaminics [8]. The second characteristic finding is facial paralysis, which can be observed in 30%-50% of patients [9]. Paralysis can be transient or permanent, or can be unilateral or bilateral.

Elias et al., in a retrospective study, found that facial edema was always present, with isolated lip involvement in 74% of cases and with only 13% of patients showing the full triad [2]. Sertace et al. concluded in a study that the diagnosis of recurrent facial palsy may not be accurate as it may be the only prominent symptom. The other two symptoms of facial edema and fissured tongue may be obscured or overlooked [1].

The differential diagnosis of MRS includes a wide variety of heterogeneous conditions. These mainly include Angioneurotic edema, lymphangioma, hemangioma, acute allergic reaction, Bell's palsy, Ascher's syndrome, neurofibroma of the lips, infectious granulomas, oral manifestations of sarcoidosis, Crohn's disease, and eosinophilic fasciitis[2, 5].

No specific biomarkers exist, and clinical diagnosis is used for exclusion. Although histopathological findings are not necessary for the diagnosis, a specimen for orofacial edema taken from the lips or skin may reveal granulomas with epithelioid cells, Langerhans type giant cells with multiple nuclei, perivascular mononuclear infiltration, non-caseating granulomas, and lymphedema [10]. These findings suggest an inflammatory etiology.

It is known that there's no definitive therapy for MRS, and therefore, recurrences are frequent [2]. Non-steroidal anti-inflammatory drugs, antihistamines with systemic and/or intralesional corticosteroids are considered as initial choices [2]. A surgical approach has been suggested only for those patients with oro-facial swelling refractory to steroid therapy and/or those present with facial disfigurement [2]. Recurrences on the same side require evaluation to rule out malignancy, particularly schwannoma [1].

The mean recurrence interval is more than one year in Bell's palsy. More than two recurrences are less frequent, and recurrences more than four are rare [1]. Recurrent ipsilateral attacks showed a worse prognosis when compared with contralateral facial palsies. The recurrence rate is higher in young patients (11-40 years). Complete resolution occurs in 70%-80% of untreated cases, and residual weakness is present in 29% of cases [10].

Recurrent Facial palsy and oro-facial swelling in children presents a diagnostic challenge. Idiopathic facial palsy remains a diagnosis of exclusion Oral cavity examination for fissured tongue is a clue and family history of recurrent facial of LMN type is important.

Recurrent facial palsy and oro-facial swelling in children present a diagnostic challenge. Idiopathic facial palsy remains a diagnosis of exclusion. An oral cavity examination for a fissured tongue is a clue, and a family history of recurrent facial palsy of the LMN type is important.

## Ethical Considerations

### Patient perspective

Overall, the patient expressed satisfaction with the work-up and the counselling provided regarding the diagnosis. However, it is important to note that future recurrences of the condition cannot be predicted.

### Informed Consent

Informed consent for the publication of the case history and accompanying photographs was obtained.

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## Conflict of interest

There are no conflicts of interest to declare in this study.

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