

Postoperative multimodal rehabilitation for spontaneous smile restoration in severe Moebius syndrome: A case report and a short literature review

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Abstract

Moebius syndrome (MS) is a rare congenital disorder characterized by bilateral facial paralysis, significantly impacting an individual's ability to convey emotions. The lengthening temporalis myoplasty (LTM) surgery is a crucial palliative intervention that can restore the smile in patients with MS by transferring the functional temporalis muscle to the paralyzed facial commissure. However, successful outcomes are closely tied to postoperative functional rehabilitation. This case report details the experience of a 29-year-old woman with severe MS who underwent LTM surgery followed by a comprehensive rehabilitation program. Over a period of 12 months, the patient achieved a spontaneous and symmetrical smile through a three-stage rehabilitation program focused on mandibular, voluntary, and spontaneous smile recovery. Speech therapy and psychological support were also integral components. This case underscores the importance of adopting a holistic approach to ensure successful and enduring outcomes from smile restoration surgery in patients with MS.

Keywords

Case report, long-term outcomes, Moebius syndrome, rehabilitation program, tendon transfer

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Introduction

Moebius syndrome (MS), a rare congenital condition, manifests with varying degrees of facial paresis, typically presenting as facial diplegia coupled with an apparent paralysis of lateral gaze.¹ Affected individuals commonly experience paresis in other muscles governed by the motor cranial nerves.² Additional deformities, such as micrognathia, talipes, and congenital agenesis of muscles, notably the pectoral group, are prevalent in this syndrome.³ A prevailing hypothesis suggests that MS may stem from dysplasia affecting either the implicated muscles or the motor nuclei of the brainstem.⁴

In a classical complete manifestation of MS, bilateral facial and abducens nerve palsy, along with a thin upper lip, are characteristic features.⁵ Facial paralysis stands out as the most conspicuous sign, impacting the ability to express emotions.⁶ The literature describes various surgical interventions, encompassing muscle transfers and nerve transposition, to restore muscle function and enhance facial expression in patients with MS.^{7–11} Notably, the success of

smile restoration surgery hinges on the implementation of a tailored rehabilitation program (RP).

This case report delves into the intricacies of postoperative rehabilitation for an adult patient with MS following bilateral lengthening temporalis myoplasty (LTM), undertaken to

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Figure 1. (a) Bilateral facial paralysis. (b) Left-hand clinodactyly. (c) Syndactyly of the fourth and fifth toes of the right foot.

recreate a spontaneous smile. The detailed account sheds light on the significance of a comprehensive rehabilitation approach in optimizing the outcomes of surgical interventions for patients with MS. We also briefly report the results of the published cases of smile restoration surgery in MS adults.

Observation

The present case was reported according to the “Case Reports” guidelines.¹²

A 29-year-old woman, diagnosed with MS since early childhood, was referred to our rehabilitation outpatient department 3 weeks postbilateral facial reanimation surgery. Prior to the recent intervention, the patient had been grappling with bilateral facial paralysis, accompanied by left-hand clinodactyly, and syndactyly of the fourth and fifth toes on the right foot (Figure 1). Notably, due to economic constraints and limited access to specialized RPs during her early years, the patient did not undergo facial paralysis surgery. The surgical approach employed for facial reanimation involved a bilateral LTM, aiming at restoring her smile and enhancing her social interactions. Upon physical examination, the patient reported experiencing spontaneous pain localized at the operation sites, radiating to the lower parts of her face. Additionally, she encountered challenges with feeding and chewing. Clinical examination revealed significant jugal and labial edema, a restricted range of mouth opening, and disruptions in facial mimicry. These findings underscore the multifaceted nature of the patient’s postoperative

challenges, emphasizing the importance of comprehensive rehabilitation tailored to address both functional and aesthetic aspects of her condition.

The RP was meticulously designed with the overarching goal of alleviating pain, reducing facial edema through manual lymphatic drainage, and addressing various facets of facial reanimation postbilateral LTM. A multifaceted approach was adopted, involving scar massage to mitigate adhesions, temporomandibular mobilization for enhanced joint motion, and a muscular approach targeting the prevention of transposed temporalis muscle degeneration while enhancing its contraction.

Following the acute phase, the long-term objectives of the RP encompassed automating the smile, sustaining voluntary joint mobility, and preserving temporalis muscle trophicity. The program extended its focus to improving swallowing, nasal breathing, and bolstering communication skills through speech therapy (ST). Central to this specialized rehabilitation approach was the meticulous restoration of the smile, integrating the transposed muscle into its new function. To ensure effective muscle neurotization, a gradual program spanning three stages for smile recovery was implemented: mandibular smile, voluntary smile, and spontaneous smile. In the mandibular smile stage, gentle mobilization of the mandibles occurred in three sectors of motion: occlusion, retropulsion, and deduction. Simultaneously, stretching exercises promoted the extensibility of the temporalis muscle. Mandibular smile acquisition typically transpired between the third- and sixth-week postsurgery, with the

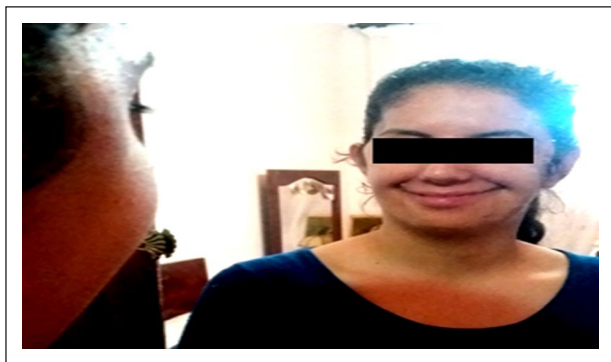


Figure 2. Spontaneous smile.

patient successfully achieving a smile as mandibular mobilization was introduced in the fifth week. The subsequent voluntary smile training stage spanned 2 months, wherein the smile was voluntarily produced through a temporalis muscle contraction, independent of mandibular mobilization. The patient, guided initially by a therapist and later self-administering, practiced bilateral and symmetric smiles by squeezing and loosening her teeth in front of a mirror for visual feedback. Ten-exercise series, repeated 10 times daily, ensured adequate practice without temporalis muscle fatigue.

By the fourth month, the patient progressed to achieving a spontaneous and symmetrical smile, without reflexive contraction or mandibular mobilization (Figure 2). Various exercises, such as reading humorous texts, raising the head, and smiling at punctuation, along with watching humorous content in front of a mirror, facilitated smile quality control. This phase represented the most challenging and time-consuming segment of the RP, extending over 6 months.

ST played a pivotal role throughout the RP, focusing on ameliorating the patient's dysarthria. At the 6-month follow-up, noteworthy improvements were observed in the production of most vowels and consonants, as well as enhanced labial occlusion. Recognizing the importance of emotional and psychological aspects in the patient's journey, specialized psychological support was concurrently provided. This support ran parallel to the smile rehabilitation efforts, aiming to assist the patient in coping with the challenge of expressing spontaneous smiles to others, a new experience for her. Additionally, she was encouraged to participate in a theater group to cultivate an emotional dimension to her smile. By the 12-month follow-up, the patient was satisfied with the result, and home-based rehabilitation practices were prescribed to further facilitate cortical adaptation of the smile. The program included regular smile exercises to be performed at home, such as reading humorous texts aloud while monitoring smile symmetry in a mirror and voluntary smile exercises to reinforce the new muscle patterns. The periodic consultations allowed the team to assess the patient's progress, provide feedback, and necessary adjustments. This holistic approach, combining in-clinic and home-based

rehabilitation, was crucial in enabling the patient to achieve a natural and enduring smile restoration.

Discussion

Through the present illustrative case, we underscore the significance of adopting a postoperative multimodal rehabilitative approach for restoring a spontaneous smile. Studying the postoperative rehabilitation effects for an adult patient with MS following bilateral LTM is crucial for at least three main reasons. First, it can provide valuable insights into the potential improvements in facial expression and overall quality of life for patients undergoing this procedure.¹ Second, it allows for a comprehensive assessment of functional outcomes such as smile symmetry, oral competence, and speech articulation, contributing to a deeper understanding of the efficacy of LTM in restoring facial function in patients with MS.¹ Third, it can identify and refine RP tailored to the specific needs of adult patients with MS undergoing LTM, leading to the development of optimized rehabilitation strategies aimed at maximizing functional outcomes and patient satisfaction.¹

The role of neuronal plasticity in MS neurorehabilitation has been highlighted in the literature.^{13,14} Ferrari et al.¹⁵ hypothesized that smile observation could improve smile recovery. The authors reported that some operated patients were able to activate the muscle graft and smile independently and voluntarily by the synchronous observation of a smile performed by another individual. Marre and Hontanilla¹⁴ described the case of a 4-year-old MS child with complete bilateral facial palsy operated with left hemiface free gracilis muscle transplant in whom a bilateral smile was noted after 8 months of rehabilitation. Moreover, Castro et al.¹⁶ reported that presurgical RP later contributed to better surgical outcomes, by enhancing the cortical representation of the structures worked upon through stimulation of sensory pathways and, probably, mirror neuron system activation. A newly recognized neurorehabilitation treatment called facial imitation treatment-synergistic activity treatment, which utilizes the presence of the mirror neuron system and the synergistic coordination between the mouth and the hand, has been proven to help achieving a more natural and spontaneous smile postsurgery and an overall improved facial functioning as compared to traditional methods.⁸ In fact, the mirror neuron system, found in the brain, is activated when an individual performs an action or observes someone else performing the same action. This system is thought to play a critical role in learning through imitation, empathy, and understanding others' actions. By using facial imitation exercises, patients stimulate their mirror neurons, which help in relearning and refining facial movements, leading to more natural and spontaneous expressions.¹⁷ There is also a well-documented synergistic relationship between the movements of the mouth and the hands. This coordination means that certain hand movements can facilitate corresponding facial

movements. By engaging the hands in specific tasks while attempting to smile, patients can enhance their ability to produce a coordinated and functional smile. This synergy is used therapeutically to reinforce neural pathways and improve motor control in facial muscles.¹⁸ Combining these elements, the treatment promotes neural plasticity, the brain's ability to reorganize itself by forming new neural connections. This is particularly beneficial postsurgery, where the brain needs to adapt to changes and re-establish motor control over the facial muscles. The repetitive and coordinated actions involving both the face and hands help solidify these new connections, leading to improved facial function and more spontaneous smiles.¹⁹ Traditional methods may focus solely on isolated facial muscle exercises. In contrast, this treatment's holistic approach engages multiple sensory and motor pathways, making the rehabilitation process more integrated and natural. This comprehensive engagement is likely why patients achieve better facial functioning and more authentic smiles compared to traditional methods.²⁰ In summary, the facial imitation treatment—synergistic activity treatment enhances facial rehabilitation by activating the mirror neuron system and utilizing the natural coordination between mouth and hand movements, fostering neural plasticity and resulting in more natural and spontaneous smiles.

It is relevant to mention that studies^{13–16} collectively highlighting the potential for a range of surgical and rehabilitative interventions report results of a pediatric population with MS and that in the case series of De Stefani et al.,⁸ only one MS adult patient (out of four) receiving gracilis transfer was included.

Few papers detailed the RP after surgical-free functional muscle transfer in adults especially after LTM, even though the surgical technique in itself is largely described in the literature.²¹ Lambert-Prou²² highlighted the significant role of ST in restoring a smile after LTM. It consists of exercises that target the muscles of the face, particularly those involved in speech and expression. These exercises help strengthen weakened muscles and improve coordination.²³ ST also promotes motor learning, where patients relearn and refine motor patterns necessary for smiling and speech. Repetitive practice of specific movements helps establish new neural pathways, facilitating more natural and controlled facial expressions.²⁴ Beyond physical exercises, ST addresses the broader aspects of communication and emotional expression. Patients practice using their new smile in social interactions, which helps them gain confidence and improve their overall quality of life.²⁵

In this report, we aimed to set up a specific, personalized, adapted, and progressive RP after LTM taking into account parameters not covered by ST. Starting postoperative rehabilitation after 3 weeks of bilateral LTM is a common practice, allowing for sufficient recovery and stabilization of the surgical site before initiating the RP.

Simultaneous performance of bilateral surgeries ensures symmetry and coordination, reduces the risk of complications

such as asymmetry, which can occur if one side recovers faster than the other, stimulates neuroplasticity, and improves functional recovery, ultimately leading to a more successful rehabilitation outcome.²⁶

The RP addressed several problems including edema, operative scars, and joint limitation affecting the temporomandibular joints, muscular tensions affecting the manducator system functioning, tongue mobility as well as swallowing and breathing problems.

Assessing and treating motor, cognitive, and psychological health impairments is crucial for comprehensive rehabilitation. Evaluating motor function is essential to understand the extent of muscle weakness, paralysis, and coordination issues. This involves detailed analysis of facial muscles, range of motion, and the ability to perform specific movements necessary for facial expressions. Targeted RP, including exercises to strengthen facial muscles, may improve coordination and restore motor function.²⁷

Cognitive assessments leading to understanding cognitive deficits helps tailor RP to the patient's needs. Therefore, cognitive training can enhance the patient's ability to participate in and benefit from motor and ST.²⁸

Psychological evaluations should identify issues such as depression, anxiety, body image concerns, and social withdrawal, which are common in patients with facial paralysis. These assessments help understand the emotional and psychological impact of the condition. Psychological interventions should include counseling, psychotherapy, and support groups to address emotional distress, enhance coping strategies, and improve mental well-being. Integrating psychological support with physical rehabilitation helps ensure a holistic approach, addressing both the mind and the body.²⁹

Even not being operated on during the childhood period, the results of combined surgery and a comprehensive RP are positive in our patient. Manktelow et al.⁹ identified that age did not affect the degree of spontaneity of smiling or the ability to smile without biting in adult patients with bilateral and unilateral facial paralysis operated with a one-stage microneurovascular transfer of a portion of the gracilis muscle. The long-term outcomes of free muscle transfer have also demonstrated sustained smile symmetry and function with the maintenance of effective muscle function and progressive improvement with time.¹⁰

In our patient, the RP has taken 12 months followed by home-based exercises to enhance cortical adaptation of the smile. It was reported that patients should maintain rehabilitation treatment for a minimum period of 30 months after which home-based rehabilitation may be considered.¹¹ We do believe that RP has to be personalized and adapted for each patient taking into account his evolution, his needs, and expectations, regardless of the length of the RP. Periodic follow-up is therefore mandatory to track the evolution and to enhance patient adherence to self-home exercises.

Moreover, there is growing interest in the potential benefits of personalized prehabilitation programs for patients

Table 1. Cases of smile restoration surgery in Moebius syndrome adults reported in the literature.

First author (year) [reference]	Number of patients	Surgical procedure	Follow up (years)	Rehabilitation program	Parameters evaluated
Manktelow (2006) ⁹	8 MS/27 (other etiologies of facial paralysis)	GMT (bilateral)	4.7	NR	Questionnaire to assess the patient's opinion of their smile Spontaneity of the smile Other functional effects of surgery
Terzis (2009) ¹⁰	1 MS/24 (other etiologies of facial paralysis)	GMT	3.5	NR	Smile quality (four independent observers rated each patient's smile using a five-category scale) Electromyography: muscle regeneration and motor units
Aguilera Salgado (2018) ¹¹	4 MS	GMT	1.5	NR	Terzis functional and aesthetic grading system Chuang's smile excursion score Questionnaire for patient satisfaction
De Stefani (2021) ⁸	1 adult/4 MS	GMT (bilateral)	NR	FIT-SAT left side Teeth clenching right side	Optoelectronic system for motion analysis
Present case	1 adult MS	Lengthening temporalis myoplasty	1	Detailed rehabilitation program	Photographs Questionnaire for patient satisfaction

GMT: Gracilis muscle transfer; FIT-SAT: facial imitation treatment-synergistic activity treatment; MS: Moebius syndrome; NR: not reported.

undergoing major surgery. These programs could be tailored to address the unique combination of motor, cognitive, and psychological impairments characteristic of each patient. By focusing on targeted interventions such as preoperative motor training, cognitive behavioral therapy, and nutritional support, tailored prehabilitation aims to optimize patient readiness for surgical interventions and improve postoperative outcomes.³⁰ Exploring personalized prehabilitation strategies represents a promising avenue for future research and clinical practice, potentially enhancing the overall effectiveness of rehabilitation efforts in this rare and challenging condition.

Table 1 details the cases⁸⁻¹¹ of smile restoration surgery in adults with MS reported in the literature.

Study limitation

In this particular case, there appears to be a potential oversight in assessing the quality of the smile and the patient's subjective evaluation of her own smile using standardized scales.

Implication for clinical care

To the best of the authors' knowledge, the existing literature lacks comprehensive information regarding the postoperative RP tailored for adult patients with MS. This case meticulously outlines a detailed RP, serving as a valuable guide for therapists in facilitating smile recovery among their adult patients with MS. It underscores the necessity for personalization and adaptation of each RP to suit individual needs.

The narrative accentuates the critical aspect of a synchronized approach between surgical intervention and rehabilitation to effectively address a spectrum of associated impairments. Notably, it emphasizes that the presented RP remains relevant and beneficial even in cases where the patient did not undergo surgical intervention during childhood, highlighting the enduring significance of such rehabilitation efforts. Further research is warranted to investigate the long-term effects of multimodal rehabilitation on smile restoration and overall well-being in patients with severe MS. While initial findings suggest promising outcomes following rehabilitation interventions, longitudinal studies are essential to assess sustained improvements in facial function, psychosocial well-being, and quality of life over extended periods. Understanding the long-term benefits of these interventions will not only validate their efficacy but also inform future therapeutic approaches aimed at enhancing the life-long outcomes of individuals living with this challenging condition.

Conclusion

This case highlights the effectiveness of multimodal and gradual rehabilitation approach following LTM for smile restoration in an adult patient with MS by integrating the transposed muscle into its new function. The automation of the smile, the main challenge of this rehabilitation, was considered as a part of a more comprehensive patient-centered RP. This holistic strategy ensuring the success of smile restoration surgery in patients with MS aims to enhance overall patient's social interactions and quality of life.

Abbreviations

MS: Moebius syndrome
 RP: rehabilitation program
 LTM: lengthening temporalis myoplasty
 ST: speech therapy

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Author contributions

S.S. conceptualized and supervised the study. C.B.M. and E.T. conducted literature review and data analysis. A.K. contributed to the rehabilitation protocol. Z.B.S.F. provided critical insights into rehabilitation interventions and patient outcomes. H.B.S. coordinated manuscript preparation and editing. All authors contributed to the drafting and critical revision of the manuscript, ensuring accuracy and clarity in the final version.

Availability of data and materials

All data showing the RP and the evolution are available.

Declaration of conflicting interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

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Ethics approval and consent to participate

The scenario pertained to standard clinical care, falling within the scope of activities exempt from ethical approval as per institutional policies. Our primary focus was on patient treatment rather than publication. When we eventually pursued publication, we ensured anonymity by de-identifying the case and excluding any patient-specific details. The accompanying literature review was conducted in a context where ethical approval was deemed unnecessary.

Consent for publication

With the intention to publish, we asked for and obtained the patient informed written consent to share her case.

Ethics approval


Our institution does not require ethical approval for reporting individual cases or case series.

Informed consent

Written informed consent was obtained from the patient(s) for their anonymized information to be published in this article.

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