

Case Report

Microstomy after Erythema Multiforme: Surgical Treatment and Results

Cristóvão Marcondes de Castro Rodrigues*, Danyella Carolyn Soares dos Reis, Luiz Gustavo Jaime Paiva, Luiz Fernando Barbosa de Paulo, and Lívia Bonjardim Lima

Department of Oral and Maxillofacial Surgery and Traumatology and Implantology Hospital de Clínicas, Federal University of Uberlândia (HC-UFU), Brazil

*Corresponding author

Cristóvão Marcondes de Castro Rodrigues, Department of Oral and Maxillofacial Surgery and Traumatology and Implantology Address: Av. Pará, 1748 - Umuarama, Uberlândia - MG, 38405-320 Block 4T Uberlândia Minas Gerais-MG, Brazil, Phone: (34) 999062400; Email: cristovao-marcondes@hotmail.com

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Keywords

- Erythema multiforme
- Diagnosis oral
- Microstomia
- Lip
- Mycoplasma infections

Abstract

Purpose: The manifestation of bullous diseases on oral mucosae leading to scarring sequelae are widely described in toxic epidermal necrolysis (TEN). These complications cause disfigurement and functional impairment. They are more rarely reported in erythema multiforme (EM). This article aims to describe a case of lip adhesion after EM induced by *Mycoplasma pneumoniae*, where corrective surgery was performed to increase the mouth opening range of a young female patient.

Methods: Through the 5-flap Z plasty, mucosal flap rotation was performed for better flexibility and better mouth opening.

Results: The interincisal distance improved by 12 mm and the intercomissural distance increased by 10 mm, with no type of scar that had an aesthetic impairment reestablishing functional improvement.

Conclusion: Through the microstomy and the choice of using 5-flap Z plasty, there was a significant increase in the patient's mouth opening, thus improving the functional and cosmetic condition, with the patient's satisfactory evolution over the long term, with no more idea relapse.

ABBREVIATIONS

TEN: Toxic Epidermal Necrolysis; EM: Erythema Multiforme

INTRODUCTION

Microstomy is a term used to describe a decreased oral opening [1]. Most cases are caused by scar contracture after facial trauma, chemical, electrical or thermal burns of the perioral tissues, excision of tumors, genetic disorders and connective tissue diseases, such as systemic sclerosis [2-5]. The cause and severity of the condition can influence the treatment approach [3]. The goals of microstomy repair include reconstructing the orbicular sphincter for proper lip function, achieving lip symmetry and well-positioned scars [6]. Different surgical and non-surgical procedures have been presented for the treatment of microstomy [7-9]. Commissuroplasty is a successful treatment modality to reconstruct the microstomy aesthetically and functionally [2,5]. In addition, several non-surgical procedures have been described to maintain adequate mouth opening by using intraoral and extraoral stretching devices [7,10]. Erythema multiforme (EM), is a rare acute mucocutaneous condition caused by a hypersensitivity reaction [4]. EM usually involves two or more mucous membranes with variable skin involvement,

it can involve internal organs and there is a 10% mortality rate for patients with extensive Stevens-Johnson syndrome [7,8]. It has been reported that EM has been triggered by several agents, particularly viruses and a variety of other infectious agents, immunological conditions, non-infectious agents, such as food additives or chemicals (benzoates and nitrobenzene) and drugs [8,9]. In this article, we report a rare case of lip microstomy after EM and discuss surgical correction and patient outcomes.

CASE PRESENTATION

A seven-year-old female patient found it difficult to open the mouth in the past six months. The mother reported that the child had erosions on the oral mucosa due to an episode of stomatitis seven months before. The mouth had become smaller and smaller by contracting the scar on the oral mucosa. Then, the patient was referred by the dentist to the oral and maxillofacial surgery service (Federal University of Uberlândia), in May 2013. The contracture of the scar was ring-shaped and located on the oral mucosa (Figure-1A, 1B and 1C). The interincisal distance was 31 mm and the distance between the commissures was 41 mm. For commissuroplasty, the surgeons used a 5-flap Z plasty, originally described by Hirshowitz et al., for stretching the contractures of the thumb mesh [11]. In the trans-surgical moment, after the

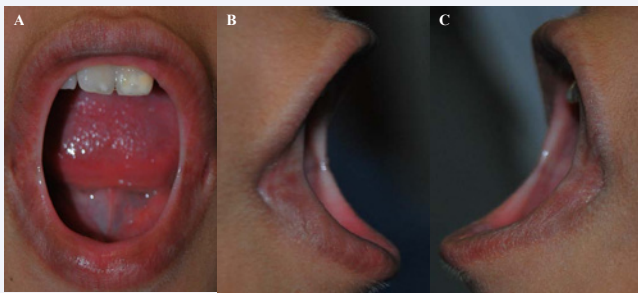


Figure 1 (A) Preoperative aspect, anterior view. (B) Preoperative appearance, right side view. (C) Preoperative appearance, left side view.



Figure 2 (A) Comparative aspect during the operation. (B) Immediate postoperative appearance.

right side had been operated, it was noted comparative difference between the commissures (Figure-2A). The flap was closed using simple sutures (Figure 2B). The postoperative period was uneventful, six months after the surgery, the interincisal distance improved 12 mm and the intercommissural distance measured 10mm. The microstomy procedure did not present a recurrence.

DISCUSSION

Erythema multiforme is a profound hypersensitivity reaction characterized by mucocutaneous lesions and ulcerative bullae; lasting normally one to six weeks, with recurrence in 25% of cases [12,13]. Within this group, it is classified as minor erythema multiforme, major erythema multiforme, Stevens-Johnson syndrome and toxic epidermal necrosis [13,14]. In this report described, the patient presented with a minor erythema multiforme induced by Mycoplasma pneumoniae with manifestation in oral mucosa, which makes it a unique case considering the non-cutaneous manifestations.

The reconstruction of the mouth with microstomy is a complex surgical procedure, in which providing a good functioning of the lips must be the first objective of the treatment method and relapses must be avoided in order to obtain stability and lasting results [15,16]. In consensus within the team, obtaining lip symmetry and an acceptable aesthetic result is the second goal of treatment.

Many procedures have been described for microstomy reconstruction in the literature as skin graft, composed of auricular lobe graft, variations of the mucous flap and a combination of skin and mucosal flaps [2,4,7,16]. In the case in question, the surgeons opted to perform a variation of the mucous flap, decreasing the

scar index as it deals with the aesthetic area, with less risk of dehiscence and familiarity with tissue manipulation.

The adoption of Z plastia of 5 flaps that did not produce skin scars during reconstruction. The polarity of the scar on the mucous flap was evenly distributed in the mouth. The aesthetically pleasing continuation of the vermilion skin and the symmetrical appearance of the neocommission was achieved, as in other reports [15,17]. Considering that this patient did not come from previous aesthetic defects, like other cases of microstomy, that usually happen in victims of burns or animal attacks, orbicular aesthetic maintenance was maintained and the range of oral functionality was maximized.

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CONFLICT OF INTEREST

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REFERENCES

1. Sato H, Toriyama K, Yagi S, Takanari K, Takama H, Sawada M, Hashimoto T, Kamei Y. Surgical correction of microstomia in a patient with antilaminin 332 mucous membrane pemphigoid. *Ann Plast Surg*. 2014; 72: 553-555.
2. Grishkevich VM. Post-burn microstomia: anatomy and elimination with trapeze-flap plasty. *Burn*. 2011; 37: 484-489.
3. Bilhan H, Geckili O, Atalay B, Arat S. Oral rehabilitation following removal of a rhabdomyosarcoma and subsequent microstomia: a case report. *J Oral Implantol*. 2011; 37: 353-360.
4. Neumann A, Coetzee PF. Freeman-Sheldon syndrome: a functional and cosmetic correction of microstomia *J Plast Reconstr Aesthet Surg*. 2009; 62: e123-e124.

5. Newkirk RE, Fomin DA, Braden MM. Erythema multiforme versus Stevens-Johnson syndrome / toxic epidermal necrolysis: subtle difference in presentation, big difference in treatment. *Mili Med.* 2020; 00, 0/0:1.
6. Kumar B, Fernandes A, Sandhu PK. Restricted mouth opening and its definitive management: A literature review. *Indian J Dent Res.* 2018; 29: 217-224.
7. Turan A, Tuncel U, Kostakog̃ lu N. The use of single rhomboid flap in reconstruction of microstomia. *Burns.* 2012; 38: e24-27.
8. Koymen R, Gulses A, Karacayli U, Aydintug YS. Treatment of microstomia with commissuroplasties and semidynamic acrylic splints. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.* 2009; 107: 503-507.
9. Sari A, Aksoy A, Basterzi Y, Unal S. Reconstruction of the oral commissure with the use of a new technique: the asterisk design. *J Craniofac Surg.* 2009; 20: 1256-1259.
10. Ayhan M, Aytug Z, Deren O, Karantinaci B, Gorgu M. An alternative treatment for postburn microstomia treatment: composite auricular lobule graft for oral commissure reconstruction. *Burns.* 2006; 32: 380-384.
11. Hirshowitz B, Karev A, Rousso M. Combined double Z-plasty and Y-V advancement for thumb web contracture. *Hand.* 1975; 7: 291-293.
12. Shi T, Chen H, Huang L, Fan H, Yang D, Zhang D, Lu G. Fatal pediatric Stevens-Johnson syndrome/toxic epidermal necrolysis: Three case reports. *Medicina (Baltimore).* 2020; 99: e19431.
13. Kara A, Devrim İ, Çağlar İ, Bayram N, Kundak S, Apa H, Altan EV. Stevens-Johnson syndrome and toxic epidermal necrolysis: a report of six cases. *Turk J Pediatr.* 2019; 61: 538-543.
14. Rifaat MA. Reconstruction of medium-sized defects of oral commissure by combining double full-thickness cheek rhomboidal flaps and a small lip switch flap. *Ann Plast Surg.* 2011; 67: 134-138.
15. Koh SH, Jeong YW, Han JJ, Jung S, Kook MS, Oh HK, Park HJ. Orbicularis oris muscle reconstruction and cheiloplasty with Z-plasty in a patient with a transverse facial cleft. *Maxillofac Plast Reconstr Surg.* 2019; 41: 55.
16. Brajon D, Bursztejn AC, Goffinet L, Schmutz JL, Barbaud A. Synéchies de la muqueuse labiale dans les suites d'un érythème polymorphe [Lip synechia after erythema multiforme. *Ann Dermatol Venereol.* 2013; 140: 291-295.
17. Kumar B, Fernandes A, Sandhu PK. Restricted mouth opening and its definitive management: A literature review. *Indian J Dent Res.* 2019; 29: 217-224.

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