

REVIEW

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Dental management of patients with microstomia. A review of the literature and update on the treatment.

Abstract: Difficulty in dental management is one of the factors that characterize the patient that requires special care in dentistry. One of the clinical conditions that make dental treatment particularly complex is microstomia. Microstomia is defined as a small and insufficient oral aperture that will hinder diagnosis and dental treatment. Although there have been reports of patients with diseases and syndromes that cause microstomia, the available literature offers only a limited number of reviews on this topic. The aim of this paper is to present a review of the etiology, clinical characteristics, diagnosis and treatment of microstomia. In addition, to describe the therapeutic adaptations to be applied in dental procedures in patients with microstomia, emphasizing the importance of a preventive approach in this group of patients.

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INTRODUCTION.

Microstomia is an insufficient or small oral aperture that, if permanent, can affect diagnosis or proper dental treatment. Adequate oral aperture is essential for facial expression, oral feeding and speech. Furthermore, it can be an obstacle for proper oral hygiene¹.

Decreased oral aperture can occur acutely caused by an infectious or odontogenic inflammatory process, but the real problem arises when it is structural or permanent. This is usually common in the group of patients with special needs².

In most cases the presence of microstomia is associated with a poor healing process of perioral soft tissues or a tissue alteration related to the temporomandibular joint².

There are causes directly related to disorders of the neuromuscular masticatory system, such as spasms, or direct damage of muscle tissue or temporomandibular joint. It also can occur following oral surgery on these structures or radiotherapy of head and neck³.

Trauma and chemical or physical burns resulting in retractable scars can alter perioral soft tissues or face tissues and lead to microstomia. It also may appear with some diseases such as epidermolysis bullosa or scleroderma and in some rare syndromes that affect connective tissue⁴.

Because there is great clinical variability in the range of oral aperture it is difficult to determine when there is a real limitation of the aperture. This would require establishing the mean value of the population by age, sex or race. Diagnosis is usually performed by measuring the interincisal distance during maximum opening of the mouth. It was established that the opening in an adult should be of at least 3-4 fingers wide⁵.

Good oral aperture is essential for a good intraoral examination, and to perform a good dental treatment. Similarly, decreased oral aperture can be an obstacle to intraoral intubation for general anesthesia, which is often necessary to perform dental treatment in patients with special needs.

Treating microstomia is complex and depends primarily on the causes that produce it. A variety of surgical procedures have been described especially when it comes to changes in soft tissues⁶; although it is also possible to use some non-surgical procedures such as oral physiotherapy exercises⁷.

The aim of this study is to review some of the dental problems caused by microstomia, adopting a preventive approach where possible and adapting techniques for the dental treatment of these patients.

ETIOLOGY OF MICROSTOMIA.

Microstomia is defined as an abnormally small oral aperture associated with several etiological factors⁸. This is a congenital or acquired condition involving a severe reduction of oral opening enough to compromise the patient's aesthetics, nutrition and quality of life⁹.

Reduced oral aperture may be caused by multiple and varied situations and may be both horizontal and vertical¹⁰. Microstomia can result from congenital defects, trauma (mechanical, chemical or thermal) or after surgical resections¹⁰.

Microstomia can be produced by a physical or chemical trauma such as electrical or chemical burns in perilabial areas that after healing leave retractable scars on these tissues, deforming and closing the corners of the lips^{1,4,11}. During the 1990s, microstomia was common in children as a result of domestic accidents, caused by electrical discharge or ingestion of caustic substances contained in cleaning products. Nowadays, due to the introduction of safety measures such accidents are rare¹².

Microstomia can also be secondary to some type of traumas or injuries after lip reconstructive surgery or after cancer surgery for head and neck tumors. In the latter microstomia may be associated with radiotherapy as an additional factor that can damage the masticatory muscles limiting their mobility^{13,14}. In some cases when the masticatory muscles or the temporomandibular joint become damaged a protective reflex contracture may occur reducing oral aperture and, sometimes, mobility can be

accompanied by pain.

Likewise, microstomia also appears as a clinical manifestation in certain systemic diseases and rare syndromes. Scleroderma is a chronic connective tissue disease characterized by changes in skin, blood vessels, skeletal muscles and internal organs¹⁵. Its causes are unknown but there seems to be an autoimmune mechanism causing an overproduction of collagen matrix, resulting in growing and hardening of connective tissue. It usually affects women between 30 and 50 years. Among its clinical manifestations we find the appearance of Raynaud's phenomenon (deformities in the fingers and toes), fibrotic skin complications, in skeletal muscles, in the gastrointestinal tract, lungs, kidneys and cardiovascular system. Involvement of the orofacial tissues is characteristic in these patients, leading to a tense and mask-like facial skin and reduced oral aperture^{7,14,16,17}.

Microstomia also occurs in patients with epidermolysis bullosa. Bullous epidermolysis or epidermolysis bullosa is an inherited condition characterized by blistering of the skin and mucous membranes as a result of trauma. It is due to the failure of anchoring elements between the layers of the epithelium and chorion. There are several types such as simple, junctional and dystrophic forms, being the latter and especially those with a recessive inheritance pattern the most severe manifestations. During clinical examination a reduction in oral aperture and numerous blisters on the oral mucosa can be observed, tongue can also appear limited in mobility and attached to the floor of the mouth. All this conditions may arise during the healing of lesions, forming scars that limit the mobility of soft tissues at various levels¹⁸⁻²¹.

Similarly, severe cases of microstomia can occur in rare syndromes such as Freeman-Sheldon with craniofacial involvement, giving the patient the appearance of a "whistling face"²²⁻²⁵. It is a form of multiple congenital contracture whose characteristics are small mouth, flat face with a small nose, long philtrum, deep-set eyes and microglossia.

There are joint contractures, abnormal hands and sco-

liosis. Its main form is autosomal dominant inheritance although there is a recessive X-linked form. Patients have a normal life expectancy and normal mental development although some cases of mental retardation have been reported. From their first year of life patients have a small oral aperture that makes feeding and oral hygiene difficult, and causes a speech delay; some patients may even suffer from respiratory system anomalies^{10,22-26}.

Another syndrome with craniofacial abnormalities and

developmental delay is the Fine-Lubinsky syndrome in which craniosynostosis, prominence of the frontal bone, flat facial profile, small nose, hearing loss and microstomia are observed. Microstomia is the most prominent manifestation making feeding difficult and entailing a risk of suffocation²⁷. Other syndromes with microstomia are Hallerman-Streiff, Schwartz-Jampel, Burton skeletal dysplasia, Leopard syndrome and Moebius syndrome^{22,28}. Table 1 summarizes the causes of chronic microstomia.

Table 1. Causes of chronic microstomia.

CAUSES OF CHRONIC MICROSTOMIA	
Trauma	Physical: direct on masticatory muscles or TMJ
	Chemical: acid or alkali burn
	Electric: electrical burn
Post cancer treatment for head and neck tumors	After cancer surgery
	After radiotherapy
Diseases or syndromes	Scleroderma
	Recessive dystrophic epidermolysis bullosa
	Freeman-Sheldon syndrome
	Fine-Lubinsky syndrome
	Hallerman-Streiff syndrome
	Schwartz-Jampel syndrome
	Burton skeletal dysplasia
	Leopard syndrome
	Moebius syndrome

DIAGNOSIS AND CLINICAL CHARACTE-RISTICS OF MICROSTOMIA.

Microstomia is characterized by a reduction in the size of the oral aperture that is not defined by specific size criteria. Diagnosis is determined by its effect on function and appearance²⁹.

In general, microstomia shows a great variability and it is difficult to establish exactly when the reduction in oral aperture has clinical significance. Normal values in the population may vary according to parameters such as age, sex or race. It is usually more frequent in men than in women and tends to decrease with age, although oral

aperture in children is smaller^{2,5}.

One of the first methods to assess microstomia was proposed by Naylor *et al.*³⁰, measuring the distance from the incisal edge of the right central incisor to the incisal edge of the right lower central incisor, or its equivalent root fragments, in maximum oral aperture. Mild microstomia is determined when the maximum oral aperture is between 41-50mm, moderate microstomia, between 31 and 40mm, and severe if the aperture does not exceed 30mm. Normal maximum oral aperture should be from 51 to 60mm¹⁸. However, it also necessary to assess soft tissue aperture, measuring the largest vertical diameter

between the upper and lower lip and the diameter between the two commissures of the mouth. The magnitude of oral aperture is a determining factor for dental diagnosis and may have implications for the treatment of these patients.

Clinically, microstomia may affect social relationships, mastication and proper oral hygiene³¹. Difficulty in oral hygiene results in a higher incidence of oral diseases such as caries, periodontal diseases or other type of oral infections that are difficult to treat due to the limited access^{11,12,16}. Likewise, it may be associated with halitosis and in certain syndromes with dysphagia, it may also be accompanied by drooling.

PREVENTION AND TREATMENT OF MICROSTOMIA.

The treatment of microstomia goes from physiotherapy to complex surgical and prosthetic treatments¹⁰.

It has been documented that tissue contractures by burn and retractable scars can be modified with pressure and splinting. An apparatus for preventing microstomia has been described (MPA: Microstomia Prevention Appliance), this has been shown effective in reducing the need for reconstructive procedures and preventing the occurrence of microstomia. It is a commissural retractor, a horizontal stretching device which opposes the contraction of scar tissue³². The MPA must be placed immediately after the facial edema has decreased and the patient must wear it throughout the day, removing it only for eating and brushing his/her teeth^{10,32}. The use of this device must be discontinued at the discretion of the physician, whose decision is based on the degree of maduration of the facial burn wound.³².

Surgical treatment of microstomia.

For the treatment of microstomia, particularly soft tissues, a variety of surgical procedures has been described. Surgical reconstruction of the oral commissures is a complex procedure because of the functional and aesthetic aspects involved in the oral cavity³³.

Procedures for the reconstruction of the commissures

(commissuroplasty) by means of upper, lateral and lower oral mucosal advancement flaps, following wedge resection of the scar tissue¹² have been described.

After surgical correction of microstomia, the tissue is replaced by fibrous connective collagen tissue. This scar tissue is subject to constant contraction for months, and together with the action of the orbicularis muscle of the lips results in the recurrence of microstomia³⁴. For the prevention and management of recurrent microstomia a compression therapy should be applied, involving a commissural splint that provides resistance to shrinkage of the scar, massaging the scar and educating the patient on exercises on horizontal, vertical and perilabial exercises¹⁰.

Sofos *et al.*³⁵ developed a new device called Whiston device used during surgery and in the early postoperative period. It acts as a surgical aid to prevent or reduce microstomia and to deal with other problems associated with facial burns. This device has a total of 4 uses; 1) acts as a circumoral and comissure retractor, 2) as a means of counter-pressure during excision of the burn, 3) counterpressure to the graft after placement, and 4) gives access to the patient's mouth to keep oral hygiene. The Whiston device is used for 1 or 3 months. Although there is only limited objective evidence for such device, it has been effective in preventing microstomia, in maintaining oral hygiene and in the maintenance of grafts³⁵.

On the other hand, surgical techniques to treat defects caused by microstomia due to the alteration of soft tissues have been undergoing several modifications over time. Triangular flaps with pedicles have been used for commissuroplasty, in the form of Z-plasty at the level of the internal mucosa or rhomboid flaps^{12,36}. Transposition of the orbicularis muscle along with an advance of the oral mucosa or free grafts from thigh or forearm have been used to rebuild the edge of the lip flaps³⁷. Flaps with oblique incision of vermilion to transpose them or bring them closer to the new oral commissure³⁷ have also been used. In recent years, a technique that uses pericomisural flaps with mucomuscular advancement, "a simple fishtail flap" to correct deformed oral commissures with the goal

of extending the oral aperture to make it functional and aesthetically acceptable without recurrence. This technique is simpler and cheaper when compared to traditional surgical methods that are generally more expensive, complex, and less practical³⁸.

Non-surgical treatment of microstomia.

Non-surgical procedures for treating microstomia are based on oral physiotherapy and the use of both static and dynamic devices to provide resistance to scar contraction or for promoting stretching and mobility respectively^{3,10}. These devices are designed to reduce scarring and keep normal functions¹⁰.

Static devices as commissural splints that keep perioral tissues in good condition during the healing period must be easy to design, relatively comfortable for the patient and effective for preventing the contraction of tissues. They are usually placed 3-6 days after the injury comprising the healing phase where fibrosis occurs^{31,39,40}.

Dynamic devices are classified into three categories according to the type of stretching provided (horizontal,

vertical, and perioral) and they are grouped according to whether the device is intraoral or extraoral, unilateral or bilateral⁴¹. Intraoral devices can be of two types: fixed or removable and both are customizable³¹. Removable devices are used because they are less expensive than fixed ones, the latter are more costly to make and require general anesthesia for placement in most cases³¹.

Patients who have suffered burns and systemic sclerosis will be good candidates for the use of stretching devices of the soft oral tissue. They are physical therapies to maintain the range of motion and minimize or prevent contractures or synechiae^{39,40}.

Other types of devices used to improve the range of oral aperture are cone-shaped corks or resins with a progressive diameter. These devices allow the patient to slowly increase the diameter of the opening⁷. They can be used at home and especially before visiting the dentist to reach maximum oral aperture⁴².

In recent years, a new alternative of non-surgical treatment for microstomia, IPL (intensed pulsed light),

Table 2. Therapeutic measures in microstomia.

THERAPEUTIC MEASURES IN MICROSTOMIA	
	Scar excision
	Commissurotomy:
SURGICAL Procedures	- Triangular flaps with pedicles (Z-plasty)
	- Rhomboid flaps
	- Oblique flap of vermillion
	- Transposition of the orbicularis muscle
	- Free grafts from thigh or forearm
NON-SURGICAL Procedures	Static intraoral devices (splints)
	Dynamic devices:
	- For stretching / mobility
	- Intraoral / extraoral
	- Horizontal / vertical / perilabial
	- Fixed / removable
	- Unilateral / bilateral
	Progressive diameter cones for progressive opening
	IPL- Intense pulsed light

most commonly known as photorejuvenation has been used. The longer wavelengths penetrate deeper into the dermis and lead to damaged collagen stimulating the formation of new collagen, which in turn results in a softer and elastic skin⁴³. Table 2 summarizes therapeutic measures used in microstomia.

THERAPEUTIC ADAPTATIONS IN PATIENTS WITH MICROSTOMIA.

A small oral aperture, large tongue or rigidity of the masticatory muscles poses a major challenge for both diagnosis and dental treatment. It has been found that performing stretching exercises and mouth opening before dental treatment increases oral aperture of a patient with microstomia in 3-5mm¹⁵. However, it is convenient to install a small rubber wedge to keep mouth space open.

Dental treatments can be uncomfortable for the patient with microstomia, and difficult or impossible to carry out by the dentist. Therefore, in these cases it is of utmost importance to establish prior prevention regimens²⁹. Avoiding cariogenic food and keeping a proper maintenance of oral hygiene is important in chronic and permanent microstomia for preventing oral disease.

For daily oral hygiene toothbrushes with small heads, short and medium-soft bristles or electric toothbrushes with small heads are recommended. Fluoride toothpaste and mouthwash rinses with 0.12% chlorhexidine and a daily application of fluoride gel with a cotton swab are also recommended in order to prevent the onset of conditions such as caries and periodontal disease⁴².

Similarly, in patients who have to undergo tumor resection surgery, followed by radiotherapy, an oral examination and all the necessary dental treatments such as the restoration of viable teeth, the extraction of teeth with active infection or poor prognosis, should be performed before the surgery²⁹.

To treat patients with microstomia, the dentist will need small rotary instruments such as dental burs with small heads and short axis^{21,42,44,45}. The use of magnifying loupes to work with more precision is often helpful.

Other materials such as cotton rolls and aspirators should also be smaller; and frequent pauses are moreover required to correctly aspirate water and saliva. ^{29,44}. To carry out restorative treatment in these patients it would be also useful to use the atraumatic restoration technique (ART), which involves making the carious cavity preparation by hand instrumentation without anesthesia or rotary instruments or aspiration, especially in back teeth²⁸. Current methods of caries prevention, such as using carbon dioxide lasers, genetically modified foods and vaccine against cavities are options with which the dentist can treat this group of patients⁴⁶.

Regarding filling materials in restorative dentistry, some authors have proposed the use of dental compomers for faster and easier insertion into the cavities under very complex conditions. However, the sealing material will depend on the achieved isolation and caries risk of the patient⁴².

Despite the use of small instruments, procedures such as endodontic treatment in molars is quite complicated, so the treatment should be done only in strategic teeth and sometimes it will be only viable in anterior teeth, being extraction the treatment of choice in the case of posterior teeth. To access the pulp chamber, sometimes dentists must use a vestibular access, and to determine the working length of the root canals the dentists will have to use the electronic apex locator due to the difficulty of taking a periapical x-ray^{29,42}.

For diagnosis, if it is not possible to take intraoral x-rays, dentists can use orthopantomography. As alternative options, it is also advisable to use small bitewing radiographs, occlusal radiograph or oblique lateral techniques⁴².

For extractions, if the extraction of all the teeth is indicated, the dentist should extract anterior teeth first to facilitate access to the back teeth⁴².

In prosthodontic treatment, making the impression represents the initial difficulty^{17,47}. It is important to adapt the different tray designs and techniques to obtain adequate impressions, since a good registry is required with minimum patient discomfort. In many cases it is

preferable to obtain preliminary impressions, followed by the use of individualized trays, modified to obtain definitive impressions^{17,40,47}. Many authors propose the use of sectioned trays to facilitate improved intraoral insertion and subsequent relocation of the sections outside the mouth^{14,17,48,49}.

Prosthesis likewise will be sectioned with hinge systems for easy placement and removal⁵⁰. There are sectioned prosthesis with various systems for relocation such as pins, orthodontic expansion screws, plastic blocks with dovetail joints, locking levers or magnets^{14,51-53}. These prosthesis are not only difficult to manufacture, they are also expensive and difficult to manage by the patient in situations where manual dexterity is compromised^{9,17}. The advantage of this design is structural durability, being more resistant to deflection and break⁵⁰. However, they have some disadvantages such as increased laboratory time, limited space for the tongue and the level of patient's engagement^{49,54}. There are other types of removable prostheses that instead of being sectioned are folding, *i.e.*, they can be folded to facilitate their placement in the mouth, making them easier to handle

by the patient⁵⁵. A folding denture in a single piece is a better choice for the functional rehabilitation of these patients. Foldable dentures may be easily introduced and easily removed from the mouth by the patient and at the same time provide adequate functionality⁵⁶.

Procedures with general anesthesia may be indicated for extensive treatments in patients with special needs. Treatment under general anesthesia allows the provision of a comprehensive dental reconstructive treatment and multiple extractions even in the presence of microstomia⁴². However, microstomia poses problems for intubation, consequently the support of an expert team becomes necessary⁴². People with microstomia may have difficulty with nasotracheal intubation³³. Patients with the Freeman-Sheldon syndrome may have problems to ventilate their airways and be at a high risk of developing pulmonary complications²⁶. Patients with the Schwartz-Jampel syndrome are at risk of developing malignant hyperthermia⁴⁵. Similarly, in Moebius syndrome there is a risk of palatine tracheomalacia and uvula weakness leading to the loss of airway function and respiratory insufficiency²⁸.

Table 3. Dental strategies for treating microstomia.

DENTAL STRATEGIES FOR TREATING MICROSTOMIA	
Dental instruments and materials	Rotary instruments with small heads or burs
	Burs with short axis
	Magnifying loupes
	Rubber wedges
	Aspirator and small cotton rolls
	Restorative material: compomers
Specific techniques	Endodontics:
	- Strategic or anterior teeth only
	- Opening by vestibular
	- Apex locator
	- Previous Orthopantomography
	Prosthesis:
	- Preliminary impressions $ ightarrow$ individualized trays.
	- Flexible memory materials
	- Sectioned prosthesis with hinges

The frequency of visits to the dentist should be based on the caries risk of the patient, every 3 or 6 months, to carry out a professional oral hygiene and the application of fluoride varnish. Dental treatment strategies for patients with microstomia are summarized in Table 3.

DISCUSSION.

Microstomia is defined as an abnormally small oral aperture that affects appropriate diagnosis and dental treatment. Microstomia is a condition that may be caused by multiple and varied situations; from birth defects, such as scleroderma, epidermolysis bullosa or in some rare syndromes that affect connective tissue⁴, or even as a consequence of the healing of the perioral soft tissues after surgical resection or trauma such as burns in perilabial areas, that while healing leave retractile scars of these tissues, deforming and closing mouth commissures^{1,4,11}.

Clinical diagnosis of this condition has not yet been clearly defined by precise size criteria²⁹, although some authors as Serrano-Martínez *et al.*¹⁸, based on the values of normal opening in adults (51 to 60mm), suggest that if the maximum oral aperture is less than 50mm we are in the presence of microstomia. However, there should be a more consolidated diagnostic method that takes into account the time of evolution of such limitation (oral aperture), to establish whether it really is a permanent condition or just a temporary trismus.

With respect to the treatment of microstomia, surgical procedures that involve the surgical reconstruction of oral commissures and non-invasive procedures such as oral physiotherapy, together with the use of static and dynamic devices have been proposed. These devices provide resistance to the retraction of the scar promoting stretching and mobility^{3,10}. However, to date, most surgical techniques to treat microstomia are complex and have a high rate of recurrence, affecting aesthetics and functionality. Consequently, developing new and innovative surgical procedures with aesthetically acceptable results together with devices that effectively prevent recurrence of microstomia is a priority.

Insufficient oral aperture can be an obstacle to maintain

proper oral hygiene and dental appearance. Dental problems are prevalent in patients with microstomia. Dental management of patients with microstomia is a challenge for the dentist and it is difficult for the dentist to perform a correct diagnosis in some dental pathologies due to the limited oral aperture. However, the problem of obtaining a proper diagnosis can be solved by the use of panoramic radiographs.

In these cases it is important to prevent oral diseases through diet, avoiding cariogenic food and keeping a proper oral hygiene, using toothbrushes with small heads and short bristles, and oral hygiene aids such as the daily use of fluoride gel and mouthwash rinses with chlorhexidine 0.12%. In cases when the dentist needs to treat a specific dental condition, the procedure should be carried out as atraumatically as possible using instruments and techniques adapted to the task. Performing mouth opening exercises before dental treatment, as well as placing a small rubber wedge for keeping the mouth space open during surgery is recommended. It would also be interesting to evaluate the prescription of a muscle relaxant to be taken by the patient one hour before the treatment.

In this review various strategies that can be taken into account for the dental treatment of patients with microstomia are described. However, information available in the literature on the topic is quite scarce, since most of the papers dealing with dental management of patients with clinical microstomia are isolated clinical cases or series of cases, with few original papers and systematic reviews of the topic.

CONCLUSIONS.

A reduced oral aperture complicates and compromises the dental treatment of patients, so dentists should have a preventive approach from an early age with proper oral hygiene techniques. This review can be useful to clarify the most important clinical aspects and familiarize oral health professionals with this condition, offering adaptive techniques for the dental treatment of patients with microstomia. Still, dental treatment in patients with microstomia is difficult, being the preventive approach the most suitable dental treatment for this group of patients.

Manejo clínico odontológico del paciente con microstomia. Una revisión de la literatura y puesta al día.

Resumen: La dificultad en el manejo odontológico es uno de los factores que definen al paciente que requiere cuidados especiales en odontología. Una de las situaciones clínicas que más dificulta el tratamiento dental es la microstomia. La microstomia se define como una apertura bucal pequeña e insuficiente que va a dificultar el diagnóstico y el tratamiento bucodental. Aunque se han descrito casos clínicos de pacientes con enfermedades y síndromes que cursan con microstomia, no hay muchas revisiones del tema en la literatura. El objetivo de este artículo es presentar una revisión sobre la etiología, la clínica, el diagnóstico y el tratamiento de la microstomia. Así como, describir las adaptaciones terapéuticas que se deben aplicar en los procedimientos dentales en el paciente con microstomia, recalcando la importancia de un enfoque preventivo en este grupo de pacientes.

Palabras clave: Microstomia, Trismus, Gestión de la práctica odontológica, Cuidado dental.

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