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Clinical case

Partial glossectomy using a stellar wedge resection technique and anterior rotation flap for the treatment of macroglossia due to lymphangioma

Glosectomía parcial con técnica de resección en cuña estelar y colgajo de rotación anterior para el tratamiento de la macroglosia por linfangioma

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ABSTRACT

Macroglossia is a condition characterized by the enlargement of the tongue's size caused by congenital malformations such as lymphangiomas, choriostomas, hamartomas, or acquired malformations such as benign or malignant neoplasms. In the mayority of cases, the treatment is surgery and can be conservative when the airway is not compromised. In this work we present the case of a 2-year-old male patient with a diagnosis of macroglossia due to lymphangioma. The treatment protocol consisted of partial glossectomy using the stellar wedge resection technique and anterior rotation flap, as well as speech therapy after the surgical phase. After a year of follow-up, persistence of the lesion was observed, meriting a second surgical intervention in which a lingual dimension of normal parameters was achieved, preserving aesthetic and functional harmony without causing neurosensory or taste alterations.

Keywords: Macroglossia, vascular malformation, partial glossectomy, lymphangioma, lymphatic malformation.

RESUMEN

La macroglosia es una afección caracterizada por el aumento del tamaño de la lengua causado por malformaciones congénitas como linfangiomas, corioestomas, hamartomas, o malformaciones adquiridas como neoplasias benignas o malignas. En la mayoría de los casos, el tratamiento es quirúrgico y puede ser conservador cuando la vía aérea no está comprometida. En este trabajo presentamos el caso de un paciente varón de dos años con diagnóstico de macroglosia por linfangioma. El protocolo de tratamiento consistió en una glosectomía parcial mediante la técnica de resección en cuña estelar y colgajo de rotación anterior, así como terapia del habla tras la fase quirúrgica. Tras un año de seguimiento, se observó la persistencia de la lesión, lo que mereció una segunda intervención quirúrgica en la que se consiguió una dimensión lingual de parámetros normales, preservando la armonía estética y funcional sin causar alteraciones neurosensoriales ni gustativas.

Palabras clave: *Macroglosia, malformación vascular, glosectomía parcial, linfangioma, malformación linfática.*

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INTRODUCTION

Sushruta-samhita in the year 600 to 300 BC. described for the first time various clinical characteristics of a patient suggestive of macroglossia, however, the first recorded definition and description of this entity dates from the 2nd century in Greece by Galen.[1] Macroglossia is a condition characterized by enlargement of the tongue caused by a wide variety of conditions, including acquired and congenital malformations such as choristomas, hamartomas, and benign or malignant neoplasms.[2] Macroglossia is usually a clinical feature in various diseases. According to Simmonds J, macroglossia has a prevalence of 4.63 per 100,000 live births, where more than half of the cases are associated with syndromes.[3] Some authors summarize the etiology of macroglossia as follows *(Table 1)*.[4-9] In the late 1990s, macroglossia was classified based on its etiology as true or false.[10] True macroglossia is secondary to histological changes that allow abnormal growth of the lingual tissue and can occur in isolation or as a manifestation of systemic conditions such as hypothyroidism, amyloidosis or in a genetic context such as Beckwith-Wiedemann syndrome or Down syndrome. Some clinical features secondary to macroglossia are summarized in the following table *(Table 2)*.[11-14]

The diagnosis of macroglossia is almost entirely clinical, however, an adequate medical history, family and pathological history are always needed, complemented with an imaging study (computed tomography, angiography and magnetic resonance imaging), laboratory studies of thyroid function, echocardiography, karyotype analysis and histopathological study in those cases in which macroglossia is caused by a pathology.[15] Lymphangiomas are a rare but existing etiology for the development of macroglossia. These are the result of an error in the embryological development of the lymphatic system, characterized by a separation of the lymphatic vessels in formation from the primitive lymphatic sacs or main lymphatic channels, developing lymphatic tissue in an abnormal location.[16] The prevalence of lymphangioma is usually 1 to 5 in 10,000 live births and is diagnosed in 90% of cases before two years of age.[17,18] According to the 2014 update, the International Society for the Study of Vascular Anomalies classifies lymphangiomas as lymphatic malformations,[19] because vascular tumors are true neoplasms with pathological proliferation, whereas vascular malformations are structural aberrations in the components of the vascular system without any evidence of pathological cell growth.[20]

Webb D.E. classifies lymphangiomas as macrocystic when the cystic spaces are greater than 2 cm, microcystic if it is less than 2 cm, or mixed. This classification has a therapeutic purpose because macrocystic lesions mostly respond to non-surgical treatment, but not microcystic lesions.[20,21] Histologically, is characterized by moderately dilated lymphatic vessels (microcystic) or macroscopic cyst-like structures (macrocystic). The lymphatic vessels diffusely infiltrate the adjacent soft tissues, showing lymphoid aggregates in the walls. The endothelium is thin and the spaces contain fluid with proteins and lymphocytes and even erythrocytes, which can be confused with mixed lesions or secondary to

Table 1: Classification of macroglossia based on its etiology

Congenital or hereditary	Acquired
Vascular Malformations	Edentulous patients
Lymphangioma	Amyloidosis
Hemangioma	Myxedema
Hemihyperplasia	Acromegaly
Cretinism	Angioedema
Beckwith – Wiedeman Syndrome	Myasthenia gravis
Down Syndrome	Lateral amyotrophic sclerosis
Duchenne muscular dystrophy	Chronic glossitis
Mucopolysaccharidosis	Postoperative edema
Type I neurfibromatosis	Odontogenic infection
Multiple endocrine neoplasia type 2B	Ranula
	Hematoma
	Lipoma
	Other carcinomas and tumor

Table 2: Clinical features and cephalometric characteristics of macroglossia.

Dentofacial defomities	Madibular prognathism
	Class III molar
	Anterior open bite
	Increased maxillary spee curve
	Transverse dimension of the maxillary and mandibular arch
	Maxillary or mandibular diastema
Respiratory disorders	Obstructive sleep apnea
Swallowing disturbances	Atypical swallowing
	Difficulty in chewing and swallowing
Alterations in phonation	Bilabial phonemes (P and B)
	Alveolar occlusive phonemes (T and D)
	Alveolar fricatives (S and Z)
Physical appearance	Retardation appearance due to hypersalivation, dyslalia, and tongue protrusion
Radiology and cephalometrics	Anterior open bite
	Mandibular or bilmaxillary dentoalveolar protrusion
	Proinclination of the maxillary and mandibular incisors
	Excessive mandibular growth with dentoalveolar protrusion
	Decreased oropharyngeal airway
	Open gonial angle
	Increased angle of the mandibular plane

hemorrhage. In intraoral tumors, lymphatic vessels are located on the epithelial surface and replace connective tissue, however, extension to deep connective tissue, bone, or muscle has also been reported.[22]

Macrocystic malformations or cystic hygroma, is a lymphatic or vascular malformation anatomically characterized by dilated lymphatic vessels due to a lack of communication between the lymphatic and venous systems.[23] They can occur anywhere in the body, in the head and neck region they can have an orbital or mediastinal extension.[24] Lymphatic malformations on the tongue are generally microcystic and have little clinical response to the use of sclerosing agents, causing even greater complications such as airway involvement, bleeding, and infection.[25] The clinical manifestations of vascular malformations are variable, since they can be focal and small, with a clinical appearance in tapioca pudding or frog eggs, or even a pearly surface due to the slightly translucent mucosa that resembles small attached vesicles. Or they may be large, causing discrete swelling, or diffuse and infiltrate large anatomic regions.[20,22] Diagnosis is 90% based on the history and clinical appearance when they are superficial and small lesions, however, deeper or more diffuse lesions may require an imaging support, which can also help to treat the lesion.[16] The main objective of the treatment is the restoration or preservation of functionality and aesthetics. The possibility of spontaneous regression in macrocystic lymphatic malformations is 3 to 15%. Therefore, observation and monitoring in asymptomatic patients is suggested.[26] Treatment can be surgical or with sclerosing agents, which are one of the first lines of treatment, sclerosing agents range from: ethanol, bleomycin, doxycycline, OK-432. However, sclerotherapy can cause serious complications such as adjacent soft tissue damage and nerve damage.[18] Macrocystic lymphatic malformations respond better to sclerotherapy than microcystic malformations.[20]

Treatment of lymphatic malformation usually requires surgery. The most appropriate management is to decrease the volume increase with neurovascular preservation of the adjacent tissue.[18] Combined treatment has also been reported, when the lesion is sought to decrease in size using sclerosing agents to move away from important neurovascular structures and then perform surgical resection.[24] The recurrence rate after complete surgical excision ranges from 0 to 27%, after partial resection from 50 to 100%, showing during the first year after excision.[18] Recurrences are common, because any residual lesion can regrow in response to trauma, infection, or hormonal changes.[20]

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CASE PRESENTATION

Next, a case of macroglossia treatment due to lymphangioma by partial glossectomy with stellar

wedge resection technique with anterior flap rotation is presented, showing so the advantages and disadvantages of the technique when compared to other types of resections. A 2-year-old male patient presented to the outpatient clinic of the oral and maxillofacial surgery service of the Peralvillo Pediatric Hospital in Mexico City. It occurs due to an increase in lingual volume of one year of evolution, limiting chewing, swallowing, speaking and breathing *(Figure 1)*. A directed physical examination revealed the presence of an anterior open bite due to macroglossia, protruding anterior teeth, lingual dorsum with inadequate hydration, dentition trauma, indentations and blood debris. The computed tomography showed a generalized tongue lesion, a decrease in the space corresponding to the airway due to an increase in volume *(Figures 2 and 3)*. A diagnosis of non-syndromic idiopathic macroglossia was given, a protocol for surgical treatment was carried out under balanced general anesthesia in the operating room.

Surgical phase: Traction was performed on the lingual vertex using a suture and lidocaine with epinephrine was infiltrated, in addition a pharyngeal plug was placed. The back and the belly of the tongue were marked off using methylene blue, the upper vertex of the star was marked 5 mm anterior to the foramen cecum and the lateral vertices were marked 15 mm from the midline, thus avoiding the lingual vessels and nerves.[27] Two divergent lines were then drawn towards the tip of the tongue and the anterior rotation flap was marked *(Figure 4)*. The wedge-shaped incision and dissection were performed in order to decrease the height of the lingual tissue, as well as the rotation of the flap at the lingual tip, preserving its shape and maintaining the papillae. At the end of hemostasis, the suture was performed through anatomical planes, thus obtaining an adequate immediate result *(Figure 5)*. One week after surgery an appointment was made for sutures removal and comprehensive evaluation. Speech therapy and pediatric orthopedic dental services were provided *(Figure 6)*. A year later, recurrence was observed, so a new surgical intervention was scheduled *(Figure 7)*.

For the second surgical phase, it was decided to use the Keyhole technique for partial glossectomy, due to the transverse volume that had to be corrected, traction was performed using sutures on the lingual tip, after which the resection was marked, lidocaine with epinephrine was infiltrated, incision was made with a scalpel and continuing with dissection using an electrosurgical unit *(Figure 8)*. Complete excision was performed and the surgical specimen was placed in 10% buffered formalin to be sent to the pathology service, where a lesion was observed rising from the proliferation of small lymphatic vessels, filled with lymph and lined by endothelial cells; These vessels were located on the superficial lamina propria of the dense fibrous connective tissue and below the hyperparakeratinized stratified squamous epithelium, in addition it was found in deeper layers mixed with the striated muscle fascicles and nervous tissue, finally the diagnosis of lymphatic malformation was given (lymphangioma) *(Figure 9)*. To evaluate the postoperative status of the patient, a one-week after surgery appointment was given for removal of stitches, and a new trainer was made by the pediatric dentistry service and speech therapy continued *(Figure 10)*. Currently the patient has an adequate evolution and improvement in terms of phonation, swallowing, occlusion and aesthetics.

DISCUSSION

Presurgical treatment

The diagnostic approach by the phoniatrics department for an associated treatment with speech therapy is essential for a correct evolution of the treatment. The speech therapy intervention should allow the patient a complete adaptation of their motor skills before 3 years of age, after this time an anatomical and habit modification becomes more difficult.[2] Orthopedic devices for functional training avoid the atypical position of the tongue and empower adequate motor coordination, showing improvement in swallowing, breathing, phonation and sucking.[28]

Conservative treatment

Conservative treatment is indicated in patients without airway involvement and without functional limitations for chewing, phonation, or swallowing; however, most patients require surgical treatment, is uncommon to see an isolated conservative therapy[2] *(Table 3)*. Small lymphatic malformations that do not affect function or aesthetics do not require treatment. Spontaneous involution has been reported in 17.7%,[29] suggesting so observation and monitoring. Palliative treatment is recommended for symptomatic lesions, coursing with pain.[24] Surgical treatment: there is controversy to determine the appropriate

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age for surgical intervention. Some authors mention that surgery must be performed before 3 years of age to obtain a better functional result,[2] however, lingual growth continues until 8 years of age,

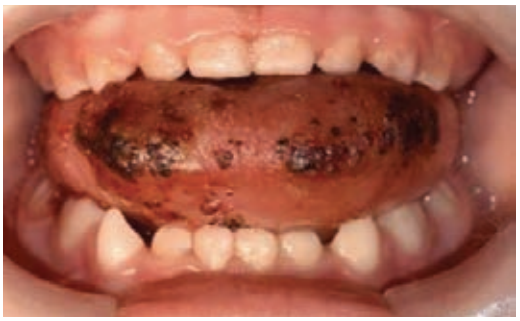

Figure 1: Generalized tongue enlargement, hemorrhagic trauma areas and blood debris.

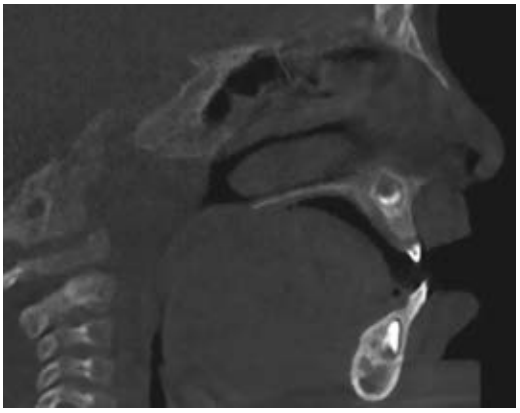

Figure 2: Computed tomography showing the airway obstruction due to the posterior lingual position.

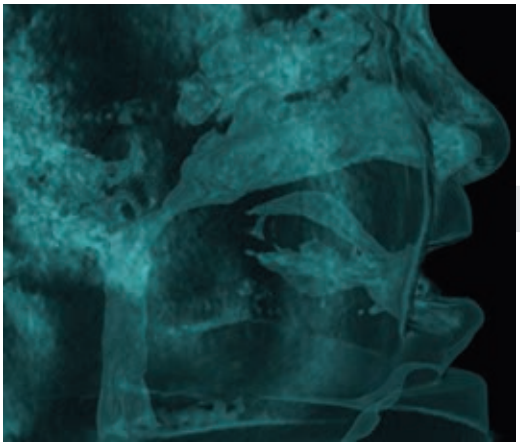

Figure 3: 3D reconstruction that shows the narrowing of the airway and the labial incompetence.

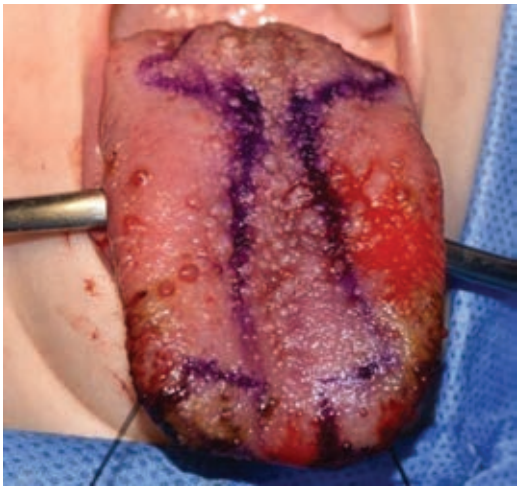

Figure 4: Surgical marking on the tongue dorsum.

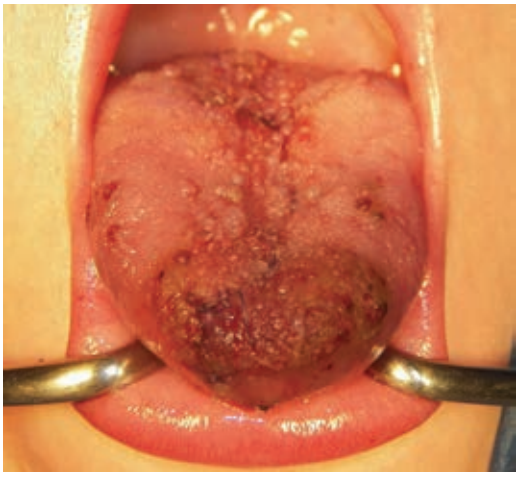

Figure 5: Surgical outcome.

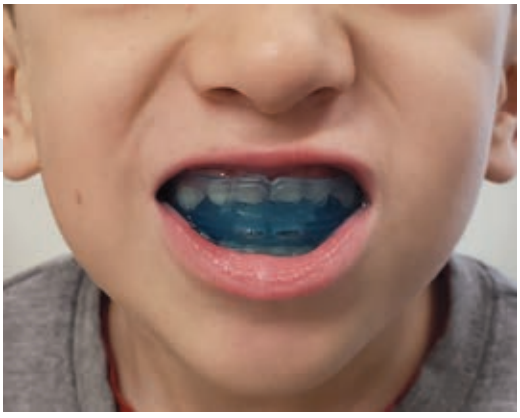

Figure 6: Training device to correct the lingual position.

during this time recurrence and reoperations are frequent.[30] Treatment for lymphatic malformations may or may not be surgical. Surgical treatment is preferred because sclerotherapy can cause edema, inflammation, infection, or airway compromise.[25]

The indications for surgical intervention are: Life-threatening due to airway restriction, feeding difficulties, word articulation misunderstanding, aesthetic concerns. Vesicles in the mucosa can be resected with laser or radiofrequency. Larger lesions can be resected when they do not respond to sclerotherapy or if complete excision can be guaranteed.[24] Surgical excision is recommended as a first instance when the diameter of the cyst in the lymphangioma is < 1 cm.[29] Partial glossectomy techniques can be classified in 6 categories: tip amputation, anterior wedge excisions, central

Figure 7: Postsurgical view after one year of treatment where clinical relapse is shown.

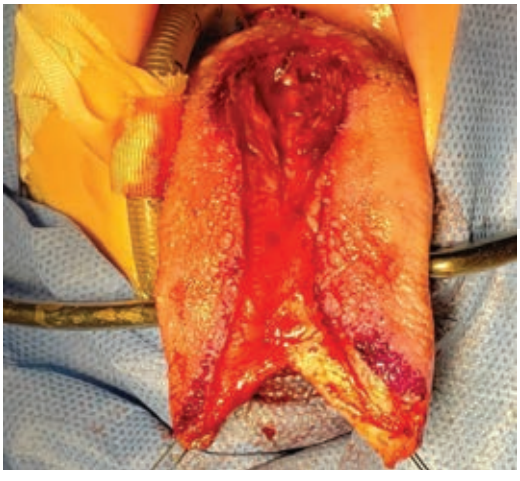

Figure 8: Keyhole Technique to perform a partial glossectomy.

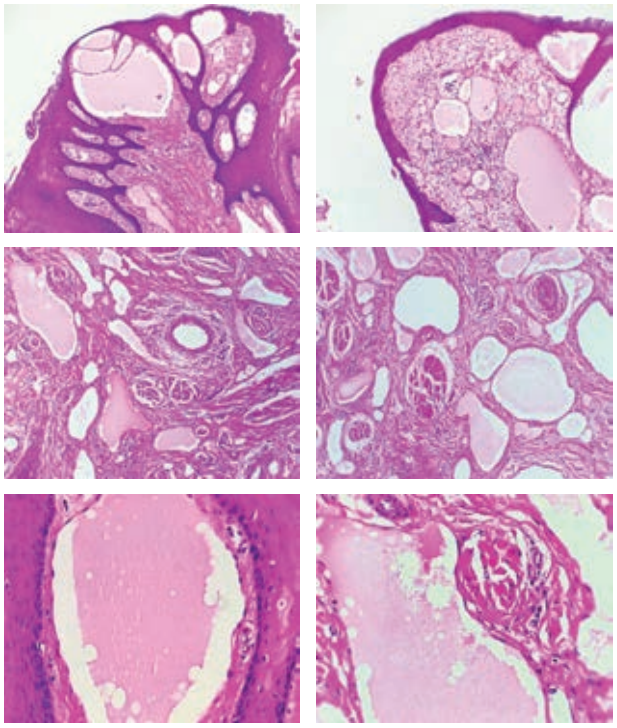

Figure 9: Histological sections in hematoxylin and eosin at 20X and 40X. The presence of subepithelial lymphatic vessels and between the striated muscle is observed.

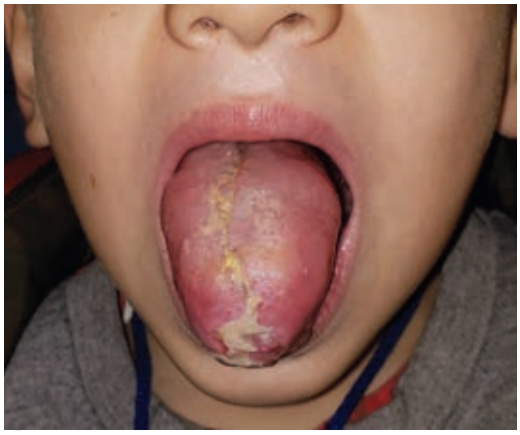

Figure 10: One week postsurgical outcome.

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reductions, dorsal flap excisions, marginal excision, and a combination of these techniques. The common goal is to achieve a normal tongue in size and position at rest, as well as the preservation of blood vessels and nerves.[31,32] Multiple resections create a sharp, asymmetrical tip of the tongue. When excising the tip, it is difficult to determine a resection without violating

Table 3: Treatment options comparative.

	Conservative treatment	Surgical treatment	Sclerosing therapy	Joint treatment
Advantages	• Spontaneous involution in 17.7% • Avoid morbidity from surgical treatment	• Improves patient function • Improves aesthetic conditions • Avoid complications related to the airway • Promotes adequate growth of the maxillomandibular complex • Recurrence of 13%	• 40% effectiveness • Mediate response of 50% and complete response of 14% • Outpatient management	• Reduces the size of the lesion and with it the damage to adjacent structures • Less recurrence compared to isolated therapy
Disadvantages	• Does not improve the patient's airway • Does not decrease the risk of complications • Requires palliative management in symptomatic lesions	• Surgical morbidity • Risk of dysgeusia and tongue paralysis • Hospitalization is needed • Risk of complications from 12 to 33%	• Does not immediately resolve complications related to the airway • Persistent pain and local fibrosis • Increases the probability of presenting anaphylactic shock	• Delayed evolution

neurovascular structures, which may result in a short tongue.[30] In this paper, the technique described by Heggie in 2013 was used to appropriately decrease the tongue in a vertical, horizontal and transverse direction, preserving the tip by means of the anterior rotation flap, thus maintaining adequate projection, function and aesthetics.[11] The success of the anterior wedge resection is due to the reduction of the central muscles without excessive manipulation reducing the loss of function. The use of this technique in the present clinical case achieved an acceptable reduction in a vertical, horizontal and longitudinal direction, as well as the preservation of the lingual tip. For the second surgical intervention, it was decided to perform a horseshoe-shaped Morgan resection, due to the recurrence of the lesion and the need to cover a greater extent of resection. This technique prevents the loss of epithelium on the tongue. It does not alter the function of the taste buds and preserves the neurovascular structures. Some authors report complete improvement nine years after surgery.[33] The anterior rotation flap allows the preservation of the lingual tip and thus the preservation of the sweet taste, however, cases have been reported in which the perception of flavors is indistinct from the amputation of the lingual tip,[34] and even when 50% of the lingual tissue is resected. Suggesting so that the perception of taste is generalized and not localized.[35,36] Among the complications of glossectomy, airway obstruction, hemorrhage, severe edema, and infection have been reported. Meriting tracheostomy, antiedema and antimicrobial management and even admission to the Intensive Care Unit.[31,35] Difficulties for articulation of phonemes after surgery have been reported in up to 4%, and a recurrence rate with surgical treatment from 15 to 53% and complications from 12 to 33%.[31,36,37] In this clinical case, the early intervention of speech therapy allowed better phonation in the pre- and postoperative stage.

Sclerosing treatment

Complete surgical resection can be difficult due to the extent and multiple lobes of the lesion, because of that sclerotherapy provides a suitable alternative to surgery. The early treatment of our patient supposed a relative urgency due to the threat to the airway caused by the lingual size, the constant trauma and the spontaneous bleeding; for this reason, sclerosing therapy was not chosen because, despite its efficacy, the results are delayed. Sclerotherapy is preferred in the early stages because it has been reported to be more effective in macrocystic lymphatic malformations. 50% of the microcystic lesions showed an intermediate response and 14% a complete response Prior to injection of sclerosing agents, the aspirated fluid must be examined to confirm the diagnosis.[26] Sclerotherapy is useful for large macrocystic lesions as it causes scarring of the cyst wall, reducing the size of the lesion, without

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causing recurrence by up to 90%.[24] Treatment using sclerosing agents allows a short hospital stay, due to its outpatient management, thus reducing the cost of treatment. When lymphangiomas have a diameter greater than 1 cm, sclerosing agents such as tetracycline, alcohol, OK-432, bleomycin, acetic acid, ethanol, doxycycline, and hypertonic saline solution can be used. In this way, damage to adjacent tissues is reduced compared to surgical treatment, and its effectiveness has been reported to be 64%. However, cases have been reported in which facial nerve damage, infection, airway obstruction, dysphonia, persistent pain, fever, local fibrosis and anaphylactic shock have been reported.[29,37]

Joint treatment

Some patients require combined treatments. In lymphatic malformations where its resection is not possible due to its anatomical location or its extension, the use of sclerosing agents initially and its subsequent surgical resection is chosen.[24,37] The recurrence rate after incomplete resection of lymphatic malformations ranges from 50 to 100%.[18] However, other authors mention that incomplete excision does not always requires reoperation.[26]

CONCLUSIONS

Primary care for patients with macroglossia must be timely and accurate, due to the impairment in their social and affective development by aesthetic and functional complications (chewing, swallowing, phonation, aesthetics) that come with it. The clinical diagnosis, the early differentiation between primary and secondary involvement, as well as the medical management of the underlying pathology, together with an adequate choice and implementation of the surgical technique, are factors that may or may not improve the patient's prognosis. Lymphatic malformations that affect the lingual tissue can cause macroglossia, with a severe degree of disturbance in a patient's quality of life, Due to its histological origin and its high recurrence, a close follow-up should be chosen, avoiding initial wide resections. The surgical technique used in this clinical case has proven to be one of the best for the treatment of macroglossia, since it can reduce both the vertical, horizontal and transverse dimensions, in addition to preserving the tip by means of the anterior rotation flap, thus maintaining an adequate morphology without sacrificing taste buds. The surgical technique is complex due to the elective dissection, however, because of this, the vascular and nervous bundle can be preserved. Therefore, the implemented technique offers excellent results in patients without severe macroglossia affection. Currently the patient does not show recurrence and has adequate phonation, chewing and respiratory function.

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