ORIGINAL ARTICLE

Epidemiology and surgical treatment in patients with lymphatic malformations

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ABSTRACT

Background: Lymphatic vascular malformations, formerly known as lymphangiomas, are alterations in the formation of lymphatic channels.
Objective: To determine the epidemiology and surgical management of patients with lymphatic malformations seen in a high-complexity pediatric hospital between 2017-2022.
Methods: The study reviews medical records in which patients with lymphatic vascular malformations with surgical resection were identified in the institution from 2017 to 2022.
Results: We included 53 pediatric patients; 52,8% were male (n=28). The tumor was the predominant sign in 96.2 % of cases (n=51), followed by respiratory stridor with dyspnoea in 7.6% (n=4). Regarding the size of the malformations, 41.5 % of cases had a size between 11 and 15 cm (n=22). The most common site was the cervical region (n=47; 88.7 %). In addition, 7 cases required preoperative tracheostomy (13,2 %), and 2 cases required postoperative tracheostomy (3,8 %). There were no complications in 79% of patients (n=42).
Conclusions: Lymphatic malformations can occur in various locations at different stages in pediatric patients, ranging in size from small masses to large tumors that can cause deformities in the head, neck, and thorax. These malformations can sometimes obstruct the airway and digestive tract, leading to significant complications.

Keywords: Lymphangioma; Lymphatic Abnormalities; Pediatrics; Head; Neck (Source: MeSH)

Epidemiología y tratamiento quirúrgico de pacientes con malformaciones linfáticas

RESUMEN

Introducción: Las malformaciones vasculares linfáticas, denominadas anteriormente linfangiomas, corresponden a alteraciones en la formación de los conductos linfáticos.
Objetivo: Determinar la epidemiología y tratamiento quirúrgico de los pacientes con malformaciones linfáticas atendidos en un hospital pediátrico de alta complejidad en el periodo del 2017-2022.
Métodos: El estudio es una revisión de historias clínicas, donde se identificaron pacientes con malformaciones vasculares linfáticas con resección quirúrgica del 2017 al 2022 en la institución.
Resultados: Incluimos 53 pacientes pediátricos, 52,8 % fueron del sexo masculino (n=28). El tumor fue el signo predominante en 96,2% de los casos (n=51), seguido del estridor respiratorio asociado con disnea en 7,6 % de los casos (n=4). En cuanto al tamaño de las malformaciones, 41,5 % de los casos tuvieron un tamaño entre 11 a 15 cm (n=22). La región cervical fue donde se localizan la mayoría de los casos (n=47; 88,7 %). Además, 7 casos requirieron traqueostomía preoperatoria (13,2 %) y 2 casos necesitaron traqueostomía postoperatoria (3,8 %). No se presentaron complicaciones en 79 % de los pacientes (n=42).
Conclusión: Las malformaciones linfáticas tienen diversas localizaciones en las diferentes etapas pediátricas con tamaños que varían desde unos centímetros hasta tumores gigantes que deforman la región de la cabeza, el cuello, el tórax, llegando incluso a presentar obstrucción de la vía aérea y digestiva.

Palabras clave: Linfangioma; Anomalías Linfáticas; Pediatría; Cabeza; Cuello (Fuente: DeCS)

INTRODUCTION

Previously called lymphangiomas, lymphatic vascular malformations are alterations in the formation of the lymphatic ducts. Lymphatic vascular malformations or lymphangiomas

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occur primarily in the pediatric age group. They are usually diagnosed in the second half of pregnancy, and in 50% of the cases, they can be present from birth and appear and develop during the patient's growth (1-3).

The body regions of greatest localization are the neck and head; the most characteristic sign is the presence of a soft, subcutaneous, painless, slow-growing tumor. However, according to certain peculiarities such as their location or size, they can also produce respiratory difficulty, dysphagia, macroglossia, oral bleeding, and exophthalmos, and they are even susceptible to present a local infection. Airway obstruction is caused by compression or displacement due to an adjacent or surrounding tumor, as well as by location in the floor of the mouth, the base of the tongue, or the hypopharynx (2).

Ultrasonography, tomography, or resonance imaging shows us the cysts' characteristics and their extension to determine a work plan. Sjogren et al. reports, according to imaging studies, that malformations in the face and oral cavity region are microcystic; in the parotid and submandibular region, they are mixed; and in the neck region, macrocystic and mixed malformations predominate (4-5).

Currently, there are more treatment options other than surgery, such as sclerotherapy or sirolimus administration and the use of laser in mucosal presentations in the oral cavity. However, there are no comparative studies but individual reports on one of the treatment modalities. In addition, there is a lack of publications to compare the results of surgery with several sessions of sclerotherapy in patients with lesions of similar characteristics and to verify the differences in the recurrence of both modalities (2).

Macrocystic lesions respond very well to surgery and sclerotherapy with statistically similar results, while mixed lesions, especially microcystic lesions, are the most difficult to remove and to sclerose, with high recurrence rates of up to 25.3%. The combination of treatments can be performed according to the decision of the parents; surgery can perform the removal of large tumors, and the residual ones receive sclerotherapy as well as sclerotherapy sessions to reduce the size of the malformation and later resection of a smaller mass (4-9).

There are several protocols for sclerotherapy, mainly for macrocystic lesions, with various substances with similar results regardless of the number of application sessions and doses. The most frequently reported complications, independent of the drug, of pain and local extravasation between 3-22% (10-12).

There are cases in which the malformations require a tracheostomy after surgery, either because of their location, or because of the post-surgical inflammatory process that reduces the airway, or because the endotracheal tube cannot be removed in the intensive care unit during the postoperative days (9).

Another difficult location to manage is the orbit since total resection is not possible or sclerotherapy is limited in its application due to the presence of microcystic lesions; in both cases, it is also due to the intimate relationship with the orbital content, thus having high recurrence rates in the management of this location. Opting for subtotal resection as a safe measure to alleviate the patient's clinical condition (13), while in other series, it is reported that sclerotherapy is the first choice for orbital location due to its safety and high effectiveness with a decrease of more than 50% of the size in all cases and improvement of visual acuity in 78.2% without compromise of visual acuity according to Barnacle et al. (14) or if there is visual compromise with hematoma aspiration, sclerotherapy as reported by Woo et al (15).

Because it is a frequent pathology in the pediatric age, being one of the national reference centers that provide treatment for the pathology, we conducted the study to determine the epidemiology and surgical treatment of patients with lymphatic malformations in patients who did not receive sclerotherapy in the institution.

We postulate that the pathology does not differentiate sex, and the presentation is primarily in preschool age; in addition, not all patients are suitable for sclerotherapy, being surgical resection is the treatment to be performed in these cases. Therefore, we aimed to determine the epidemiology and surgical treatment of patients with lymphatic malformations seen in a high-complexity pediatric hospital from 2017-2022.

METHOD

Design

This is a cross-sectional study.

Participants

Based on the review of medical records, patients with lymphatic vascular malformations in a high-complexity pediatric hospital were identified, of which all patients who received surgical treatment from January 2017 to December 2022 with anatomopathological results of lymphatic malformation, cystic lymphangioma, cystic hygroma, lymphangiohemangioma or hemangiolymphangioma were included.

44 patient records were excluded: 6 whose pathology did not correspond to that of the study, 04 with insufficient data for case registration, and 34 who underwent sclerotherapy as the only treatment.

Variables of interest

The variables of age, sex, location of the malformation, signs and symptoms, size, treatment, complications, recurrences, and sequelae were identified.

Analysis plan

The statistical analysis was performed using SPSS (Statistical Package for Social Science) version 25. The mean, minimum, maximum, median, absolute, and relative frequencies were analyzed for the descriptive analysis.

Ethical aspects

The study is based on the fundamental ethical principles according to the Declaration of Helsinki, such as nonmaleficence and confidentiality. The information collected was absolutely confidential and exclusively for the study. Authorization was obtained for the review of clinical history data and consent. In addition, the study was approved by the Ethics Committee of the Instituto Nacional de Salud del Niño San Borja.

RESULTS

87 patients with lymphatic vascular malformations were identified in the period under study, of which 53 (60.9 %) cases received surgical treatment at the institution, and the other 34 (39.1 %) cases received sclerotherapy treatment. Surgical treatment was performed on those patients who were not candidates for sclerotherapy because they were microcystic or mixed, patients who did not want sclerotherapy treatment, or patients under 1 year of age.

In total, information was collected on 53 pediatric patients who underwent surgery. There was a homogeneous distribution with 28 (52.8 %) males and 25 (47.2 %) females; the age group with more cases was between 1 to 5 years, with 25 (47.2 %) patients, followed by the group between 1 to 11 months with 15 (28.3 %) cases, having an average of 2,9 years and a range of 6 days to 15 years for all patients.

It is essential to mention that 94.3 % of the patients had no therapeutic history except for two patients who had previous surgery and another case who received both sclerotherapy and tracheostomy in another institution (see Table 1).

The most striking and prevalent sign among patients was a visible and palpable tumor in 51 (96.2 %) cases, followed by respiratory stridor associated with dyspnea in 4 (7.6 %) patients (see Figure 1).

Based on the size, we took as reference the largest diameter of the malformations evaluated according to the graphic scale of 5 cm and its progression every 5 cm in the images by tomography or resonance, having mainly 22 (41.5 %) cases between 11 to 15 cm; with an average value in the size of 10.9 cm for the total of the study. Based on the imaging studies, 35 (66 %) cases had mixed lesions (macrocystic and microcystic), 11 (20.8 %) cases had only macrocystic, and 7 (13.2 %) cases had microcystic images. As for their location, the cervical region was where they presented in the vast majority of patients, 47 (88.7%) cases. In the extracervical presentation, there were 4 (7.5 %) cases with extension to the mediastinum, four with involvement of the floor of the mouth, 3 (5.7 %) cases with lingual involvement, and three at the parotid level.

According to the Serres classification for cervical lymphatic malformations, the unilateral supra- and infra-hyoid location with 21 (44.7 %) cases was the most common, followed by the unilateral supra-hyoid with 12 (25.5 %) cases.

Although tracheostomy was not necessary in most patients, seven cases (13.2 %) required preoperative tracheostomy, and

Table 1. Patient characteristics (n=53)

Features	Group		n	%
From the patient	Sex *	Female	25	47,2
		Male	28	52,8
	Age *	Neonate	6	11,3
		1 - 11 months	15	28,3
		1 - 5 years	25	47,2
		6 - 10 years	2	3,8
		11-15 years	5	9,4
	Therapeutic Background *	None	50	94,3
		Previous surgery same institution	1	1,9
		Previous surgery at another institution	1	1,9
		Sclerotherapy and tracheostomy	1	1,9
Of the lymphatic malformations	Clínical *	Tumor	51	96,2
		Dyspnea and respiratory stridor	4	7,6
		Dysphagia	2	3,8
		Tongue bleeding	1	1,9
	Size *	Less than 6 cm	4	7,5
		6 - 10 cm	18	34,0
		11 - 15 cm	22	41,5
		16 - 20 cm	6	11,3
		21 - 25 cm	1	1,9
		25 - 30 cm	1	1,9
		30 - 35 cm	1	1,9
	Location *	Neck	47	88,7
		Mediastinum	4	7,5
		Mouth floor	4	7,5
		Tongue	3	5,7
		Parotid	3	5,7
		Arm pit	1	1,9
		Fronto-orbital	1	1,9
		Genial	1	1,9
		Occipital	1	1,9
		Thoracic	1	1,9
	Ranking of Serres**	Supra and infrahyoid unilateral	21	44,7
		Suprahyoid unilateral	12	22,5
		Infrahyoid unilateral	7	14,9
		Supra and infrahyoid bilateral	5	10,6
		Infrahyoid bilateral	1	2,1
		Suprahyoid bilateral	1	2,1

***Note**: * n=53 ** n=47*

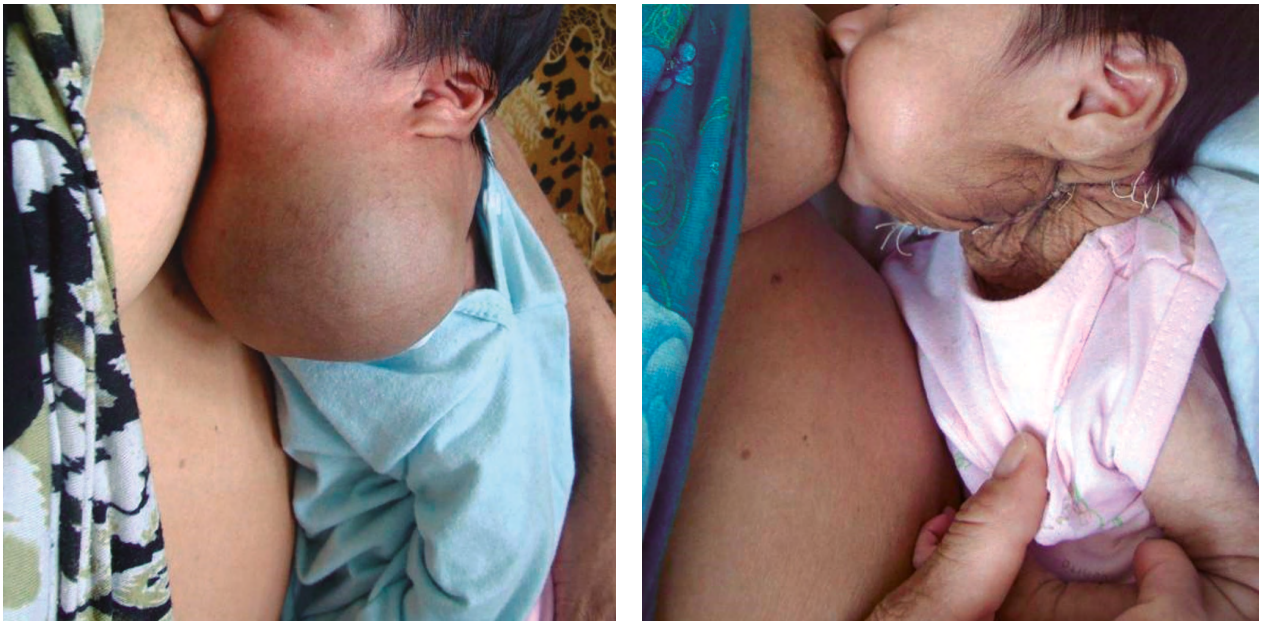

Figure 1. Unilateral supra and infrahyoid lymphangioma, pre and postoperative photo.

Note: Patient with cervical tumor corresponding to unilateral supra and infra hyoid lymphatic malformation, pre (left) and postoperative image (right)

two cases (3.8 %) required postoperative tracheostomy, which was performed in a period of 15 to 30 days due to extubation with respiratory stridor and tugging that increased in frequency and intensity (see Table 2).

Wide resection was performed without evidence of residual tissue in 44 (83 %) cases and partial resection in 9 (17 %) cases due to the location of the malformation in areas of essential structures. Of note, 42 (79.2 %) patients had no complications; however, labial paresis, thoracic duct fistula, and presentation of a seroma (requiring drainage and compressive bandage) occurred in 3

Table 2. Characteristics of tracheostomies performed

Preoperative tracheostomy	N°	Age	Location	Size
Tracheostomy removal	1	11 years	Tongue	9 cm
		08 days	Bilateral supra and infrahyoid	22 cm
		08 days	Unilateral supra and infrahyoid	20 cm
		10 days	Bilateral Supra and infrahyoid and tongue	14 cm
Continued tracheostomy	6	06 months	Bilateral Supra- and infrahyoid, tongue and floor of mouth	12 cm
		08 months	Unilateral supra and infrahyoid	11 cm
		01 year	Unilateral supra- and infrahyoid and thoracic	35 cm
Postoperative tracheostomy	**N°**	**Age**	**Location**	**Size**
Intubación prolongada	1	02 months	Unilateral supra- and infrahyoid and mediastinal	15 cm
Parálisis de cuerda vocal unilateral por lesión nervio recurrente	1	03 months	Bilateral infrahyoid	30 cm

(5.7 %) cases for each. However, vocal cord paralysis due to recurrent nerve injury occurred in a different case. One patient died of septic shock due to Pseudomonas aeruginosa as a postoperative complication (see Table 3).

Table 3. Characteristics of complications presented (n=53)

Complications	N°	%	Age	Location	Size	Treatment
None	42	79,2				
Labial paresis	3	5,7	08 days	Supra and infrahyoid	20 cm	Parenteral nutrition, Physical medicine for 10 days, Healing
			01 year	Supra- and infrahyoid and thoracic	35 cm	Parenteral nutrition, Physical medicine for 10 days, Healing
			14 years	Supra and infrahyoid and genial	10 cm	Parenteral nutrition, Physical medicine for 10 days, Healing
Thoracic duct fistula	3	5,7	06 days	Supra and infrahyoid	12 cm	Ondansetron 10 days
			10 days	Supra- and infrahyoid and mediastinum	16 cm	Ondansetron 15 days
			06 months	Supra- and infrahyoid and mediastinum	14 cm	Ondansetron 10 days
Seroma	3	5,7	01 month	Supra and infrahyoid unilateral	15 cm	Surgical cleaning, healing and compression
			01 year	Unilateral infrahyoid	9 cm	Surgical cleaning, healing and compression
			09 years	Unilateral suprahyoid	6 cm	Surgical cleaning, healing and compression
Septic shock, death	1	1,9	02 months	Unilateral supra- and infrahyoid and mediastinal	20 cm	Antibiotic coverage, tracheostomy, thoracic and cervical drainage
Recurrent nerve palsy	1	1,9	03 months	Bilateral infrahyoid	12 cm	Tracheostomy

DISCUSSION

Lymphatic malformations present mainly in the pediatric age; our study shows that 86.8% of the cases were presented in children under five years of age, with 13.2% in children older than 6 years of age and without finding statistically significant differences in terms of presentation according to sex, results similar to those described by Waner et al (2). In addition, Sjogren et al. (4) mentioned that 90% of the cases are in children under 5 years of age, and 75% are in the head and neck region.

The visible and palpable tumor was the predominant sign in the population, and according to the larger size of the malformation and the younger age of the patient, dyspnea with respiratory stridor was another striking sign in 7.5 % of the cases. With different sizes, 75.5 % of the cases presented malformations with a larger diameter between 6 to 15 cm, without leaving aside diameters between 16 to 20 cm (11.3 %). The 20.8 % of the malformations were of macrocystic architecture; however, due to the age, clinical condition of the patient, location of the malformation, or choice of the parents, surgical treatment was determined. In addition, unilateral cervical presentation is the predominant one, with 85.1 % of the cases. According to international publications (1-3), the locations are very diverse, usually with macrocystic and microcystic components, which present asymptomatic tumors as the main characteristic. Sjogren et al. (4) state that there is no consensus on the laterality; even so, there are series with incidence between 52-70 % of cases on the left side.

Total resection of the lymphatic malformation is not always possible; in our study, 17% of the cases had a partial resection of the tumor due to the proximity and compromise of adjacent structures since the possibility of its removal increases the risk of morbidity. Waner et al. (2) indicate that macrocystic tumors respond well to surgery. In contrast, microcystic tumors have a high possibility of recurrence, while García et al. (3) mention that the orofacial location has a significantly higher recurrence rate than the cervical location. In our series, only 3 (5.7 %) cases had a history of previous surgical interventions, while 94.3 % were followed up at 6, 12, and 24 months without recurrence.

Tracheostomy was required in 50 % of the operated neonates due to the difficulty in orotracheal intubation. In comparison, it was only required in 20 % of the cases younger than one year of age, all with malformations larger than 10 cm in greatest diameter. The other case with preoperative tracheostomy corresponded to macroglossia, which made intubation impossible. On the tenth postoperative day, the tracheostomy was removed with decreased local edema. Bagrodia et al. (6) mention that early tracheostomy for relief of airway obstruction is sometimes necessary, even without complete surgical resection.

The involvement of the airway by lymphatic malformations is due to mechanical obstruction when the tongue and neck region are involved by displacement or extrinsic compression of the larynx or trachea. At the same time, neonatal or infant patients may even present laryngomalacia or tracheomalacia.

Because of this, they are usually patients classified as difficult intubation, which is why tracheostomy should always be the final alternative to failure of orotracheal intubation with the support of equipment or even flexible fibroscopy.

Postoperative edema, the risk of injury to the inferior laryngeal or pneumogastric nerves, and laryngomalacia or tracheomalacia are described in the literature as factors that influence the integrity of the airway, which is why our two cases that required tracheostomy were due to prolonged

intubation in the patient with sepsis and vocal cord paralysis with stridor and respiratory distress.

According to García et al. (3), complications reach up to 35%, with operative site infections followed by functional alterations in the VII, IX, X, XI, XII, laryngeal or phrenic nerves. In our series, 79.2 % did not present postoperative complications, and in the patients who did, one case died (1.9 %) due to persistent septic shock; the case developed mediastinitis, which required broad-spectrum antibiotic treatment and drainage procedures in both cervical and thoracic. Regarding labial paresis, he received physical therapy, recovering with physical therapy; it occurred in tumors more significant than 10 cm in diameter. The thoracic duct fistula required treatment with intravenous ondansetron, total parenteral nutrition, cures, and bandage, resolving in 10-15 days. The seromas presented required drainage in the operating room, curation sessions, and bandages to resolve completely in 8-10 days. The recurrent nerve paralysis was evidenced postoperatively, and after 1 month of control and with audible stridor and respiratory distress, laryngoscopy was performed, evidencing the vocal cord paralysis, being one of these cases that required tracheostomy after the procedure.

Both surgery and sclerotherapy are presented in case series, with a preference for one technique as the first choice treatment, according to the author. However, a superior treatment modality between surgery and sclerotherapy has not yet been defined for macrocystic lesions and is less effective for microcystic lesions (5-8). The recommendation for future studies is to confront two groups, one surgical and the other with complete sclerotherapy, to evaluate the success and recurrence rate between the use of sclerotherapy and to recommend studies to evaluate the success rate through these techniques in the future.

Conclusion

Lymphatic malformations present a variety of locations and sizes in the pediatric population, ranging from small lesions to giant tumors that can deform areas such as the head, neck, and thorax and, in some cases, cause airway and digestive tract obstruction. Although sclerotherapy, which requires multiple infiltration sessions, has been established as an effective treatment, surgery remains a vital option, especially for those patients with contraindications to sclerotherapy or whose parents choose not to use it. The authors suggest surgery as the preferred treatment in patients with respiratory compromise younger than one year. At the same time, in other cases, they recommend postponing surgery unless the patient's symptomatology intensifies significantly.

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