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Clinical Letter

Mediastinal Venous-Lymphatic Malformations: Thoracoscopic Resection

To the Director,

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The lymphangiohemangiomas (LAH) is a rare disease first described in 1983 by Angtuaco et al.^{1–3} The LAH are currently called venous-lymphatic malformations (VLM).^{1,2,4} The VLM are considered tumor-like vascular dysplasias that contain vascular and lymphatic tissue.^{2–5}

Our aim is to present a clinical case of VLM, perform a review of the current literature and describe the surgical technique.

A 53-year-old female patient with a personal pathological history of a Type 1 diabetes mellitus and social tabaquism consult to our center due to the discovery of a mediastinal cystic tumor in the routine clinical controls. Clinically asymptomatic, she didn't present dysphagia, cough, dyspnea, dysphonia or any symptoms of vascular compression. The physical examination and laboratory blood test was normal. A thoracic computed tomography (CT) scan with intravenous contrast show a hypodense mediastinal cyst of 73 mm \times 46 mm \times 28 mm (Fig. 1A).

The surgical team decided to perform a thoracoscopic (VATS) resection of mediastinal tumor. During the surgery, an ultrasonic scalpel was used to avoid heat dissipation and to achieve a correct

C D

Fig. 1. (A) Thoracic coronal CT scan with intravenous contrast show a hypodense mediastinal cyst (red circle) with 9 HU. (B) Mediastinal cyst surrounded by the superior vena cava and the right phrenic nerve (yellow circle). (C) Microscopic image \times 40, lymphatic follicles (thin red arrow) and vascular structures (thick yellow arrow). (D) Image \times 80, lymphatic follicles (thin red arrow).

sealing of the sectioned structures. Special care was taken in the dissection to preserve the integrity of the cyst and the structures surrounding it: Medial – vena cava, phrenic nerve and recurrent laryngeal nerve; lower – azygos vein; posterior – thoracic trachea and vagus nerve; superior – right subclavian vein (Fig. 1B). The dissection began by releasing the cyst from the vena cava and phrenic nerve, subsequently releasing it from the azygos vein and finally from the trachea and subclavian vein. The excision of the specimen was performed with an endobag through the working port. The patient's postoperative course was uneventful. The anatomopatological study reveals a lymphoid tissue with venous vascular proliferation, compatible with LAH (Fig. 1C, D).

LAH represent between 0.7 and 4.5% of all mediastinal tumors and usually appear in childhood. The histogenesis of vascular malformations is not clear, although VLM have been considered to result from aberration in vasculogenesis.^{2,3} Patients with VLM had a range age of 3–60 years and a mean age of 26.4 years.² The VLM are rarely found in the mediastinum,² but the anterior mediastinum is one of the most common sites for lymphangiohemangiomas.^{2–4}

The symptoms depend on the location of the tumor. Patients may have cough, chest pain, hemoptysis, venous varicosities in the neck, and dysphasia because local compresion. Other patients may be asymptomatic.^{1–3}

The imagenological characteristic of mediastinal VLM depends on the proportions of lymphatic and venous components. MR could be useful for elucidating slow-flow components of VLM. The CT angiography is excellent for delineating the VLM and their relationships with periferal structures. A LAH should be included in the list of differential diagnoses of an incidentally found low-attenuating mediastinal mass. 3-3,5

The diagnosis of VLM can be suspected by imaging, 2 and is finally confirmed by the anatomopathological study of the surgical specimen. $^{2-5}$

The surgical resection is the treatment choice of the patients with VLM. 1,2,4,5 A follow-up is indicated after resective surgery because the recurrence of mediastinal VLM were noted in case of incomplete resection, but have not been reported after complete excision. 2,3

We present this case because there are few published cases of VLM due to its low incidence. This article presents a form of treatment with a thoracoscopic approach. We believe that resective surgery with free margins (RO) is the best treatment for VLM and the VATS approach is the most appropriate for this type of mediastinal tumors, given the excellent visualization of the cyst to be able to perform RO surgery, without damaging the surrounding anatomical structures. Currently, there are no guides and/or protocols for the treatment of this pathology and taking into account the small number of patients presented in the literature, we believe that it is important to present this study since it shows the method of adequate minimally invasive resection without recurrence.

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Contribution of each author

- JG Yaryura Montero: collected and analyzed the data, wrote the most of the paper.
 - RA Benavidez: data interpretation, conceived the ideas of the study.
 - MA Cafaro: data interpretation, head of the project, manager.

Conflicts of interest

The authors declare not to have any conflicts of interest that may be considered to influence directly or indirectly the content of the manuscript.

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