literature. Dermatol Surg. 2018;44:1247–61. http://dx.doi.org/10.1097/DSS.0000000000001566.
6. Monten C, Berwouts J, Veldeman L, Creytens D, Braems G. A case of eccrine porocarcinoma, located in the breast: the pitfalls reviewed. Med Case Rep. 2017;4(S1):004. http://dx.doi.org/10.21767/2471-8041.S1-004.

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Bilateral chylothorax and chylous ascites resulting from the spontaneous rupture of a retroperitoneal lymphangioma☆

Quilotórax bilateral y ascitis quilosa como consecuencia de la rotura espontánea de un linfangioma retroperitoneal

Injury to the lymphatic system, either by obstruction or by traumatic disruption, gives rise to leakage of lymphatic fluid that can accumulate in the thoracic and abdominal cavities.[1] Chylothorax is the most common cause of pleural effusion in neonates, although in adults it represents only 3% of cases of pleural effusion. Chylous ascites is even less common, with an incidence of approximately 1 in every 20,000 cases. The simultaneous accumulation of lymph in the serous cavities is very rare. It is usually associated with non-traumatic etiologies[2] and can lead to severe nutritional deficiency as well as immunosuppression that could lead to life-threatening situations for the patient.[3]

We present the case of a 38-year-old patient with no relevant personal history who started with an episode of progressive dyspnea associated with massive left pleural effusion compatible with chylothorax. Initially, he responded to conservative treatment with pleural drainage, *nil pero os*, and total parenteral nutrition. The extension study to determine its origin included a thoracoabdominal computed tomography scan that showed an incidental finding of a lesion in segment V of the liver. Liver segmentectomy was performed, and the pathology study reported benign hepatic adenoma. The immediate postoperative period was uneventful. However, 7 days after hospital discharge, the patient was readmitted due to predominantly left bilateral pleural effusion compatible with bilateral chylothorax. Since the discharge through the left drain was > 500 mL/day and taking into account the anatomical variations of the thoracic duct described in the bibliography,[4] fluorescence-guided left video-assisted thoracoscopy and ligation of the thoracic duct were performed after administration of indocyanine green as a localization method. After surgery, the discharge of the left hemithorax decreased considerably; however, the right side increased to more than one liter a day, so it was decided to surgically address the right side, this time using lateral thoracotomy: the thoracic duct was located and tied at that level. As a result of this last intervention, the collected discharge from both thoracic drains fell significantly (<100 mL/day). Despite this, on the second postoperative day, the patient developed abdominal distension and pain along with oliguria and impaired renal function. Abdominal ultrasound revealed an abundant amount of free fluid that, after aspiration and drainage, again showed the presence of lymph.

Once all the possible secondary medical etiologies potentially causing this condition had been ruled out, magnetic resonance lymphography was performed, which detected an abdominal mass with a craniocaudal diameter of 11 cm that had not been observed on the initial computed tomography scan. The lesion extended from the celiac vessels up to the renal hila surrounding the abdominal aorta in early phases (Fig. 1), with extravasation of the contrast agent into the abdominal cavity in late phases (Fig. 2).

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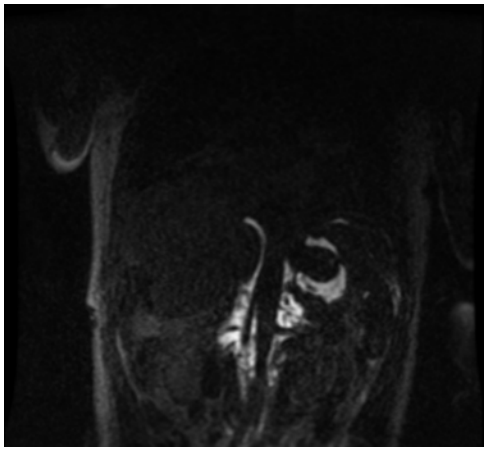

Fig. 1 – Early-phase lymphangiography showing an abdominal mass measuring 7 cm that extends from the celiac vessels to the renal hila surrounding the abdominal aorta.

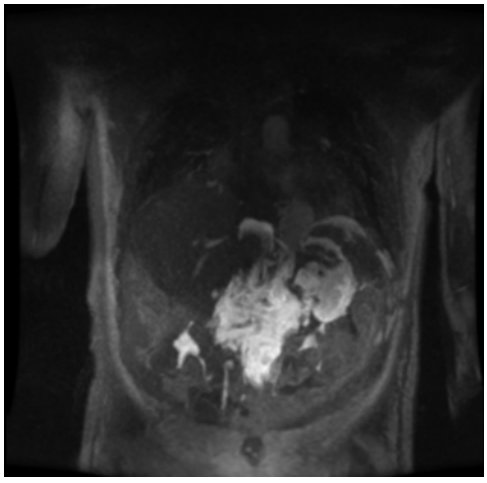

Fig. 2 – Late-phase lymphangiography showing extravasation of the contrast agent towards the abdominal cavity.

The clinical-radiological diagnosis of bilateral chylothorax was established, in association with chylous ascites resulting from spontaneous rupture of a retroperitoneal lymphangioma.

Since the therapeutic options from the surgical point of view were limited and based on few clinical cases described in the literature,[5] immunosuppressive treatment with sirolimus was initiated. The amount of discharge decreased, but not enough for withdrawal of the drainage tubes or to consider the condition resolved. Therefore, also based on references with low scientific evidence,[1,3] we opted for low-dose adjuvant radiotherapy (10 Gy, 1 Gy/day) directed at the retroperitoneal mass. Afterwards, the patient progressed favorably, with progressive withdrawal of the drainage tubes 5 days after the end of radiotherapy treatment. Since discharge, the patient has maintained intermittent immunosuppressive treatment and a diet rich in medium-chain triglycerides, presenting no new episodes.

Lymphatic malformations are rare benign anomalies that result from defective embryonic development of the primary lymphatic structures, which generally present as dilations of the lymphatic vessels, creating multiple cysts that vary in size.[4] Retroperitoneal lymphangiomas account for less than 1% of all lymphangiomas; they are usually asymptomatic and are diagnosed incidentally.[6] The presentation as bilateral chylothorax or chylous ascites is extremely rare; there are published case reports of chylothorax[7] or chylous ascites[8] as an isolated manifestation of mediastinal-retroperitoneal lymphangiomas. However, the simultaneous presentation of both entities in a lymphangioma of retroperitoneal location alone has not been described previously. Magnetic resonance lymphography after intranodal administration (bilateral inguinal lymph nodes) of 50% gadolinium is the most appropriate technique for visualization and mapping of the lymphatic vascular system. In all cases, the therapeutic decision must be individualized and based on the type of malformation, size, location, and associated symptoms. Classically, surgery has been considered the treatment of choice, but resection is often incomplete and associated with high rates of recurrence. For this reason, less invasive therapeutic options have recently been described, such as sclerotherapy, laser treatment, radiotherapy and pharmacological treatment.[1,5,9,10]

REFERENCES

1. Brown S, Abana C, Hammad H, Brown A, Mhlanga J, Binder C, et al. Low-dose radiation therapy is an effective treatment for refractory postoperative chylous ascites: a case report. Pract Radiat Oncol. 2019;9:153–7.
2. Kashyap A, Mahajan V, Whig J, Gupta S. Bilateral chylothorax, chylopericardium and chylous ascites. Lung India. 2011;28:133–5.
3. Johnson D, Klazynski P, Gordon W, Russell D. mediastinal lymphangioma and chylothorax: the role of radiotherapy. Ann Thorac Surg. 1986;41:325–8.
4. Amodeo I, Colnaghi M, Raffaeli G, Cavallaro G, Ciralli F, Gangi S, et al. The use of sirolimus in the treatment of giant cystic lymphangioma: four case reports and update of medical therapy. Medicine (Baltimore). 2017;96:e8871.
5. Ernemann U, Kramer U, Miller S, Bisdas S, Rebmann H, Breuninger H, et al. Current concepts in the classification, diagnosis and treatment of vascular anomalies. Eur J Radiol. 2010;75:2–11.
6. Rani DV, Srilakshmi R, Malathi S, Raghupathy V, Bagdi RK. Unusual presentation of a retroperitoneal lymphangioma. Indian J Pediatr. 2006;73:617–8.
7. Wagenhäuser PJ, Mölleken C, Walter JW, Heyer CM, Nicolas V. A combined mediastinal and retroperitoneal cystic lymphangioma as a rare cause of recurrent pleural effusion. Pneumologie. 2013;67:265–9.
8. Hermanutz KD, Boldt I, Frotscher U. Chylous reflux and chylous ascites in lymphatic dysplasia, with the lymphographic demonstration of mediastinal and retroperitoneal lymphatic cysts. Rofo. 1975;122:14–9.
9. Churchill P, Otal D, Pemberton J, Ali A, Flageole H, Walton JM. Sclerotherapy for lymphatic malformations in children: a scoping review. J Pediatr Surg. 2011;46:912–22.

10. Railan D, Parlette EC, Uebelhoer NS, Rohrer TE. Laser treatment of vascular lesions. Clin Dermatol. 2006;24:8–15.

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Acute Massive Idiopathic Gastric Ischemia☆

Isquemia gástrica masiva aguda de causa idiopática

The stomach is one of the most vascularized abdominal viscera in the organism. For this reason, gastric ischemia is considered a rare condition. There are known causes of gastric ischemia associated with toxic, mechanical factors, vascular damages or infections. Nevertheless, sometimes there is no clear trigger. Massive idiopathic gastric ischemia is an extremely infrequent entity with very few cases described in the medical literature.[1]

We present a case of patient with massive idiopathic gastric ischemia.

A 61-year-old woman with a past medical history of hipertension and mastectomy with axillary lymphadenectomy due to a lobular carcinoma of the breast three years ago, presented to the emergency department for abdominal pain of 48 h evolution. On clinical examination, the patient had hypothermia (34.7 °C), a blood pressure of 96/50 mmHg, pallor, obnubilation and tachycardia (150 min^{-1}). Marked rebound tenderness was found on abdominal examination. Laboratory values showed 940 leukocytes/mm^3 (88.3% neutrophils), a C-reactive protein level of 48 mg/dl, a procalcitonin level of 48 mg/dl and a venous pH was 7.30. After initial evaluation and resuscitation, the patient underwent a thoracic and abdominal CT scans that identified bilateral pleural effusion, free intra-abdominal fluid and mucosal edema in the gastric antrum and gastric body. Gastroscopy revealed ischemic mucosa in the cardia. Through a midline laparotomy, a massive gastric necrosis was found (Fig. 1) and total gastrectomy was performed. During the procedure, the patient was hemodynamically unstable and required vasoactive drugs at high doses. For these reasons, primary anastomosis was not performed and an esophagostomy and a feeding jejunostomy were created. Pathologic examination revealed an extensive necrosis, with focal transmural involvement that caused subtotal mucosal hemorrhage, severe congestion of the entire wall and acute necrotizing inflammation that extended throughout the submucosa, reaching muscle and focally subserosa and serosa. After surgery, the patient was admitted to the intensive care unit. However, the septic shock was refractory to the measures and the patient died at 14 hours postoperatively. The family rejected autopsy.

The most frequent causes of gastric ischemia are volvulus, intrathoracic herniation of the stomach, and massive gastric dilatation due to mechanical factors such as intestinal obstruction, pyloric stenosis or atonicity of the stomach caused by anorexia nervosa and electrolyte imbalance.[1,2] On the other hand, infectious causes (necrotizing gastritis) have been reported, generally involving immunocompromised patients (diabetes, AIDS, neoplasia).[3] The intake of toxins such as caustics can also cause necrosis in the stomach.[1] In our case, mechanical obstruction and vascular compromise have been ruled out at CT findings. In the same way, the intake of toxins was discarded through a meticulous anamnesis. Bacterial growth was not evident in the analysis of the piece as the cause of the disease. For these reasons, the etiology of this massive gastric ischemia was considered idiopathic.

Generally, the most common clinical manifestations are abdominal pain, emesis or those derived from a complication, such as intestinal obstruction or perforation. Physical findings can be abdominal distension, tympanism and tenderness.[3]

Currently, therapeutic options in gastric ischemia include nasogastric tuve, to decompress the stomach, and immediate surgery. When we suspect a gastric ischemia, surgical therapy

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