tolerated. Follow-up CT showed clear radiological improvement, with practically complete resolution of the metastatic pleural component and reduced effusion, disappearance of the right parahiliar mass, no new lesions in the lung parenchyma, and resolution of the masses in paracardiac fat. Hepatic metastases were no longer visible in the abdomen, with improvement of ascites and peritoneal carcinomatosis. However, persistent bone involvement and pelvic mass were observed, although considerably decreased in size.

Krukenberg tumor is malignant metastasis to the ovary of mucosecretory signet-ring adenocarcinoma that usually originates from a gastrointestinal tumor and more rarely from other sources. It represents 1%–2% of all ovarian tumors.[1,2] Gastric and colorectal cancers collectively account for almost 90% of cases.[1] Other less common primary sites are breast, appendix, small intestine and endometrium. Lung cancer rarely causes ovarian metastases (scarcely 2%–4% of cases).[3,4] Lung tumors that metastasize to the ovary are most often small cell disease (up to 40%–45% of cases).[3–5] The exact mechanism of metastatic spread to the ovaries is unknown.[1,6] The most common histopathological finding is adenocarcinoma with signet-ring mucosecretory cells. Clinically, patients may be asymptomatic or have non-specific symptoms depending on tumor involvement.[1,2,6] Ascites may be observed in up to 39% of cases,[6] as was the case in our patient. Despite the fact that studies have been performed to determine the typical immunohistochemical profile of this type of involvement, no single expression has been defined due to widely varying patterns.[7] Nevertheless, napsin-A is a useful marker for differentiating primary lung adenocarcinomas from adenocarcinomas originating in other organs, and, in conjunction with TTF-1, increases the sensitivity and specificity of tests identifying the origin of metastatic lung adenocarcinomas.[4] No specific therapeutic strategy has yet been defined. If the primary tumor is detected, it should be treated according to histology and staging.[6] Prognosis is poor due to the metastatic nature of the disease, and mean survival is 14 months.[1]

In conclusion, we report the case of a Krukenberg tumor originating from lung adenocarcinoma, a very rare presentation in lung cancers, but one that should be taken into account in the differential diagnosis of ovarian masses.

References

1. Aziz M, Kasi A. Cancer, Krukenberg Tumor. Treasure Island: StatPearls Publishing; 2018.
2. Tamayo T, Santana O, Fiallo L. Krukenberg tumor. Rev Cubana Obstet Ginecol. 2014;40:96–101.
3. Rojas B, Carazo B, Ania A, Guardia L, Arribas T, Florián J, et al. Metástasis ováricas de carcinoma de células pequeñas de pulmón. Rev Chil Obstet Ginecol. 2013;78:240–3.
4. Losito NS, Scaffa C, Cantile M, Botti G, Costanzo R, Manna A, et al. Lung cáncer diagnosis on ovary mass: a case report. J Ovarian Res. 2013;6:34, http://dx.doi.org/10.1186/1757-2215-6-34.
5. Irving JA, Young RH. Lung carcinoma metastatic to the ovary: a clinicopathological study of 32 cases emphasizing their morphologic spectrum and problems in differential diagnosis. Am J Surg Pathol. 2005;29:997–1006.
6. Kubeček O, Laco J, Špaček J, Petera J, Kopecký J, Kubečková A, et al. The pathogenesis, diagnosis, and management of metastatic tumors to the ovary: a comprehensive review. Clin Exp Metastasis. 2017;34:295–307, http://dx.doi.org/10.1007/s10585-017-9856-8.
7. Crăciun MI, Domşa I. Immunohistochemical diagnosis of Krukenberg tumors. Rom J Morphol Embryol. 2017;58:845–9.

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Chylothorax Due to Venous Thrombosis in a Double-Lung Transplant Patient☆

Quilotórax secundario a trombosis venosa en portador de trasplante bipulmonar

To the Editor,

Chylothorax is defined as the extravasation of chyle from the thoracic duct or another lymph vessel to the pleural space, due to injury or rupture. The most common cause is traumatic, while most non-traumatic chylothorax is caused by neoplastic disease (mainly lymphoma).[1,2]

Other causes have been described, including infections, systemic diseases (sarcoidosis, lupus, etc.), jugular vein thrombosis, etc. Central venous thrombosis (CVT) produces increased pressure in the thoracic duct that can cause it to rupture, with leakage of chyle into the pleural cavity. This is a rare event, associated in most cases with thrombotic complications of central venous catheters.[3,4] In other cases, CVTs may be due to prothrombotic diseases, such as cancer, inflammatory diseases, and chronic infections.[3]

The diagnosis of chylothorax is established when the level of triglycerides in pleural fluid is greater than 110 mg/dl; values of less than 50 mg/dl exclude the diagnosis, while for values between 50 and 110 mg/dl, the diagnosis is confirmed by the presence of chylomicrons.[4,5]

The importance of this disease lies in the fact that chyle is very rich in nutrients (fat, fat-soluble vitamins, proteins) and also contains cells (mainly T lymphocytes). If left untreated, therefore, it can cause immunodeficiency, severe malnutrition, and coagulopathy, etc., and may even be life-threatening.[4]

We report the case of a 61-year-old man, recipient of a double-lung transplant due to chronic obstructive pulmonary disease (COPD) in April 2017, whose initial progress was satisfactory. Eight months after transplantation, he was admitted to hospital for cytomegalovirus replication in plasma with 5039 copies/ml. Skin lesions were also observed on both legs, and had been appearing in outbreaks for the past 2 weeks. Skin punch biopsy was performed, with the following result: "Leukocytoclastic vasculitis. Skin punch showing epidermis with discrete, significant histologic changes. The superficial dermis shows neutrophilic infiltrate surrounding vascular structures associated with endothelial necrosis, extravasation of red blood cells, and karyorrhexis". Eleven months

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after surgery, the patient presented a decline in forced expiratory volume in 1 second (FEV_1) of 1 liter associated with decreased vesicular breath sounds in the right hemithorax on pulmonary auscultation, so he was admitted for further study. Chest X-ray on admission showed a new right pleural effusion. The study of pleural fluid obtained by thoracentesis found a milky fluid suggestive of chylothorax, so during the same procedure a 20 F chest drain was placed, with collection of 2500 ml of liquid. Biochemistry of the fluid showed triglycerides 2462 mg/dl. Cervical-chest-abdominal computed tomography (CT) revealed the presence of intraluminal thrombi of acute appearance in the right subclavian vein and left accessory jugular vein, with chronic thrombosis of the left jugular vein.

Standard treatment for chylothorax began, with chest drain, total fasting, complete parenteral nutrition[4] and subcutaneous octreotide[6–8] for a period of 14 days, in addition to initiating anticoagulation with low molecular weight heparin at a dose of 1 mg/kg body weight. Four days after reintroducing an oral fat-free diet, the milky fluid reappeared, so conservative treatment began again for a period of 12 days. It returned again after restarting oral intake, so surgical treatment was indicated.

Surgery consisted of the identification and ligation of the thoracic duct and its accessory branches[4] by video-assisted thoracic surgery (VATS). Initial progress was good, but at 48 h, a massive right hemothorax developed that required surgical reintervention with polytransfusion of blood products and admission to the post-surgical recovery unit. During the postoperative period, the patient developed acute renal failure that required an adjustment of the dose of tacrolimus. Two weeks after surgery, a clinical picture emerged consistent with acute rejection, so treatment began with corticosteroid boluses of 500 mg/12 h for 3 days, with satisfactory resolution of the syndrome.

Coinciding with the high-dose corticosteroid treatment, the patient developed a surgical wound infection, which was treated by negative pressure therapy and subsequent surgical closure. There was no relapse of the chylothorax. He was finally discharged in a satisfactory manner.

Thrombosis of the left jugular vein as a cause of chylothorax is an extremely rare entity, according to the literature reviewed.[9–11] However, this entity has not previously been reported following lung transplantation.

The most important cause of CVT is related to catheters, but other causes such as systemic diseases have also been described. In our case, the patient did not undergo central venous manipulation. Eight months post-transplantation, he presented cytomegalovirus (CMV) replication in plasma, coinciding with the development of leukocytoclastic vasculitis[12] treated with oral prednisone at doses of up to 60 mg/day.

In leukocytoclastic vasculitis, vascular injury occurs due to the deposit of circulating immune complexes, which activate the endothelial cells, the fibrinolytic process, and the complement cascade that can lead to intravascular thrombosis.[12] Some types of vasculitis, such as Behçet's disease, are known to be related to deep vein thromboses,[13,14] so the presence of vasculitis in an immunosuppressed, polymedicated patient could have a role in the pathogenesis of the CVT that caused the chylothorax.

Anticoagulation is the mainstay of treatment for central venous thrombosis, and plays an important role in cases of CVT-related chylothorax when vessel recanalization is achieved.[3,15] In our case, anticoagulation was administered, but the expected recanalization was not achieved after one month of conservative treatment, so surgery was performed.

Lung-transplant patients present various complications, such as those described in this case, so rare associations of multiple nosological entities that may be derived from immunosuppression or from infectious states must be ruled out. These patients are extremely complex and pose a diagnostic and therapeutic challenge.

References

1. García J, Alemán C, Jáuregui A, Vázquez A, Persiva O, Fernández de Sevilla T. Quilotórax en adultos. Revisión de la literatura a partir de una serie de 17 casos. Arch Bronconeumol. 2017;53:407–8.
2. Álvarez Vega P, Cadenas Menéndez S, Sánchez Serrano A, Fernández Sánchez JL, Martín Sánchez MJ, López Zubizarreta M. Quilotórax secundarios a trombosis del sistema venoso superior. Arch Bronconeumol. 2017;53:83–4.
3. Kho SS, Tie ST, Chan SK, Yong MC, Chai SL, Voon PJ. Chylothorax and central vein thrombosis, an under-recognized association: a case series. Respirol Case Rep. 2017;5:e00221.
4. Nair SK, Petko M, Hayward MP. Aetiology and management of chylothorax in adults. Eur J Cardiothorac Surg. 2007;32:362–9.
5. Maldonado F, Hawkins FJ, Daniels CE, Doerr CH, Decker PA, Ryu JH. Pleural fluid characteristics of chylothorax. Mayo Clin Proc. 2009;84:129–33.
6. Ismail NA, Gordon J, Dunning J. The use of octreotide in the treatment of chylothorax following cardiothoracic surgery. Interact Cardiovasc Thorac Surg. 2015;20:848–54.
7. Bellini C, Cabano R, de Angelis LC, Bellini T, Calevo MG, Gandullia P, et al. Octreotide for congenital and acquired chylothorax in newborns: a systematic review. J Paediatr Child Health. 2018;54:840–7.
8. Yin R, Zhang R, Wang J, Yuan L, Hu L, Jiang S, et al. Effects of somatostatin/octreotide treatment in neonates with congenital chylothorax. Medicine (Baltimore). 2017;96:e7594.
9. Isik Y, Binici O, Kaiti I. Chylothorax developing due to thrombosis in the subclavian vein. Euro J Gen Med. 2013;10:243–5.
10. Wu ET, Takeuchi M, Imanaka H, Higuchi T, Kagisaki K. Chylothorax as a complication of innominate vein thrombosis induced by a peripherally inserted central catheter. Anaesthesia. 2006;61:584–6.
11. Van Veldhuizen PJ, Taylor S. Chylothorax: a complication of a left subclavian vein thrombosis. Am J Clin Oncol. 1996;19:99–101.
12. Pulido-Pérez A, Avilés-Izquierdo JA, Suárez Fernández R. Vasculitis cutáneas. Actas Dermosifiliogr. 2012;103:179–91.
13. Belkouch A, Belyamani L. Jugular vein thrombosis due to Behçet disease. Pan Afr Med J. 2015;20:216.
14. Zhang L, Zu N, Lin B, Wang G. Chylothorax and chylopericardium in Behcet's diseases: case report and literature review. Clin Rheumatol. 2013;32:1107–11.
15. Siu SL, Yang JY, Hui JP, Li RC, Cheng VY, Cheung TW, et al. Chylothorax secondary to catheter related thrombosis successfully treated with heparin. J Paediatr Child Health. 2012;48:E105–7.

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