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Cystic Metastasis of a Giant Cell Tumor Causing Recurrent Spontaneous Pneumothorax

Metástasis quística de un tumor de células gigantes causante de neumotórax espontáneo recurrente

Dear Editor:

A 17-year-old male was admitted to the Emergency Department with cough and episodes of hemoptysis. The patient had a history of a giant cell tumor (GCT) in the left tibia, resected 6 months previously. Chest computed tomography (CT) revealed pulmonary nodules, some of which were cavitated (Fig. 1A). Laboratory test findings were unremarkable. The patient's sputum was negative for acid-fast bacilli. He was referred for fiberoptic bronchoscopy with bronchoalveolar lavage. The bronchoalveolar lavage fluid contained a small amount of blood, and was negative for neoplastic cells. Cultures were negative for fungus and bacteria. Video-assisted thoracoscopy was performed, and the biopsy findings from one of the nodules were compatible with GCT metastasis. The patient started a new chemotherapy cycle. Four months later, he had an episode of chest pain associated with hemoptysis. A new CT examination showed a left pneumothorax, and cavitated thick-walled nodules with ground-glass halos (Fig. 1B and C). The pneumothorax was drained. The patient evolved well, with pulmonary re-expansion. Eight months later, he had a new episode of chest pain and dyspnea. CT showed a spontaneous left pneumothorax, and evolution of the cavitated nodules into thin-walled cysts (Fig. 1D). In this phase, the patient presented metastasis to intraabdominal lymph nodes in addition to the pulmonary metastases. He underwent new chest drainage and pleuroscopy with bilateral pleurodesis through the intrapleural instillation of talc. During pleuroscopy, pleural metastases were detected. The patient underwent a chemotherapy regimen with six cycles of doxorubicin and cisplatin, which

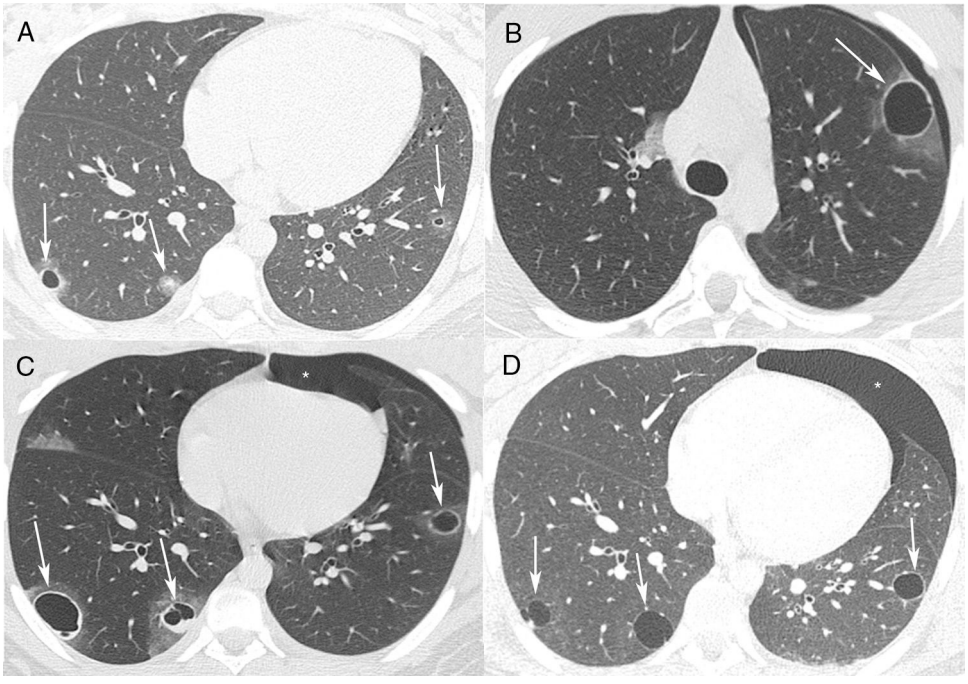

Fig. 1. (A) Axial chest CT with pulmonary window settings shows bilateral small pulmonary nodules, two of which are cavitated (arrows). (B, C) CT performed 4 months later demonstrates a left spontaneous pneumothorax (asterisk) and growth of the nodules, which now present with relatively thick walls and ground-glass halos (arrows). (D) CT performed 1 year after A shows a new left pneumothorax (asterisk) and evolution of the cavitated nodules into thin-walled cystic lesions (arrows).

resulted in regression of some of the lung lesions. He remains in outpatient follow-up with no new complication 1 year after the last pneumothorax.

GCT in the bone is a primary intramedullary tumor; it is generally benign, but can be locally aggressive and even metastatic. Malignant transformation and distant metastasis are extremely uncommon. Malignant transformation may occur as a result of dedifferentiation of the primary tumor or secondary to previous radiation therapy. Metastasis of GCTs most commonly arises in the lungs. Pulmonary metastases are more likely to appear in patients with recurrent GCTs, and often have an indolent course; they are rarely fatal.[1,2] Cavitation of metastases is extremely rare. Pulmonary metastasis initially presents as a solid mass, with an air-filled cavity formed after discharge of the necrotic material inside. The wall of a cavitated metastasis is generally thick and irregular, although thin-walled cavities can be found and may be seen with other lesions at various stages of excavation. The exact mechanism of cavitation is usually difficult to determine, but the cause is presumed to be tumor necrosis or a check-valve mechanism that develops by means of tumor infiltration into the bronchial structure. A potential complication of cystic metastases is pneumothorax, caused by necrosis of subpleural metastases producing a bronchopleural fistula.[3,4] The use of pleurodesis with talc in the treatment of pneumothorax associated with cystic pulmonary metastases has been described in the literature.[5,6] Authors[7,8] have suggested that given the high rates of recurrence, pleurodesis should be performed after the first spontaneous pneumothorax in patients with diffuse cystic lung diseases, rather than waiting for a recurrent episode. To prevent possible right pneumothorax, our patient underwent prophylactic resection of subpleural metastatic lesions and bilateral pleurodesis. The long-term prognosis and survival rate are favorable for patients with pulmonary metastasis, except for those with sarcomatous transformation, who have a worse prognosis.[1,2]

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Nódulos pulmonares cavitados con relación a la enfermedad por depósito de IgG4

Cavitary Lung Nodules Associated with IgG4-Deposition Disease

Estimado Director:

La enfermedad relacionada con IgG4 es un trastorno multisistémico caracterizado por la formación de lesiones fibroinflamatorias, provocando el fallo funcional de los tejidos afectados[1].

Las 2 características principales de esta enfermedad son la infiltración tisular por IgG4 y su elevación sérica, no presente en todos los pacientes[2].

La enfermedad relacionada con IgG4 supone un reto diagnóstico ya que imita muchos otros procesos incluyendo neoplasias, infecciones y enfermedades autoinmunes. De ahí la necesidad de realizar biopsias a los tejidos afectados en la mayoría de los casos para llegar al diagnóstico.

Presentamos el caso de una mujer de 79 años, exfumadora, asintomática, remitida a nuestro servicio para estudio de nódulos pulmonares, hallazgo casual en una radiografía de tórax realizada para un estudio preoperatorio. La fibrobroncoscopia no mostró alteraciones significativas. Se realizó una tomografía computarizada (TC) de tórax (fig. 1) en el cual se visualizaron 2 lesiones de morfología irregular gruesa con amplia cavitación central y calcificaciones groseras periféricas sin contenido en su interior, en ambos segmentos apicales de 45 y 40 mm de diámetro en lóbulo superior derecho (LSD) y lóbulo superior izquierdo (LSI), respectivamente.

La analítica de sangre presentaba únicamente anticuerpos antinucleares (ANA) positivo 1/160 granular con cuantificación normal de IgG4 (53 mg/dl).

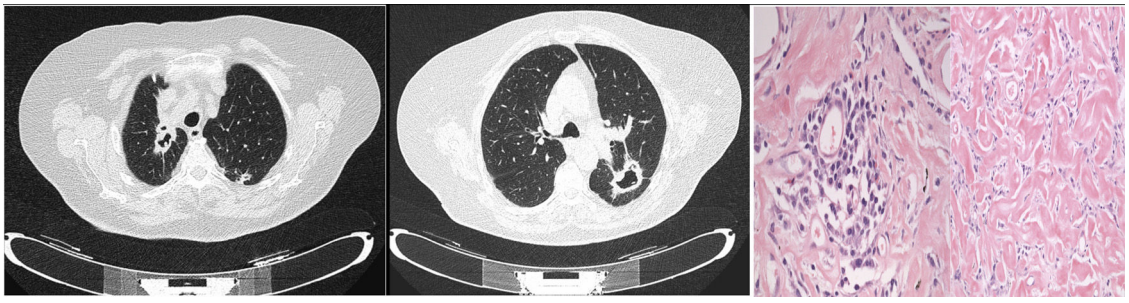

Figura 1. Pruebas de imagen y anatomía patológica.