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Scientific Letters

Muscle Involvement in Long-term Malignant Pleural Mesothelioma[☆]



Afectación muscular de un mesotelioma maligno pleural de larga evolución

To the Editor,

We report the case of a 60-year-old patient, active smoker (40 pack-years), employed in the construction sector. In 2011, he presented in the emergency room with atypical chest pain. Chest X-ray revealed significant pleural effusion, and he was hospitalized. The study was completed with a computed tomography (CT) of the chest and abdomen, which showed bilateral pleural thickening with significant pleural effusion. Positron emission tomography (PET)-CT confirmed bilateral pleural uptake but ruled out pulmonary nodules and other distant involvement. Tumor disease was suspected, so a pleural biopsy was obtained using videoassisted thoracoscopy, and pleural malignant mesothelioma was confirmed. The extension of the lesion rendered it inoperable, and the patient was referred to medical oncology. Here, we began firstline treatment with a platinum and pemetrexed doublet, and partial response according to RECIST criteria was recorded after 4 cycles, with reduction of the pleural implants and effusion, and low toxicity. Given the clear clinical benefit, treatment continued with single-agent pemetrexed as maintenance therapy with radiological follow-up every 3 months. 1 The disease remained stable with optimal oncological control for 2.5 years, when the patient developed pain in the region of the left thigh. Examination revealed increased consistency in the area, which was hard and painful on palpation. A magnetic resonance imaging study performed to rule out a neoplastic process in this region showed a soft tissue lesion involving the whole of the thigh, extending to the gluteus. Radiological characteristics were consistent with secondary involvement (Fig. 1). Given the rarity of the observation, an ultrasound-guided biopsy was performed, confirming metastatic malignant mesothelioma. In view of our patient's prolonged oligometastatic disease, local radiation therapy to the muscle lesion was proposed. Moreover, because he was clearly benefitting from systemic treatment and in view of the lack of second-line options, he continued the same therapeutic regimen with oncological monitoring for another 12 months.

Our case is an example of the clinical and therapeutic management of long-term pleural malignant mesothelioma which raises 2 points of interest: prolonged survival achieved with the treatment, and the presence of secondary musculoskeletal involvement. Malignant mesothelioma is a rare tumor that is characterized by involvement of the serous membranes, most commonly the pleura. It is traditionally associated with exposure to asbestos, although

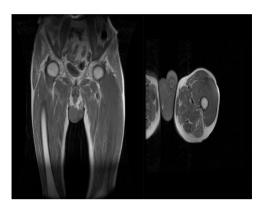


Fig. 1. A musculoskeletal MRI images: on the left, a coronal slice and on the right a sagittal slice, showing the extension of tumor.

other risk factors have been identified.^{2,3} The initial clinical presentation is characterized by pleuritic pain and pleural effusion. Diagnosis is difficult, and depends in many cases on ruling out other entities. Pleural biopsy is required for a definitive diagnosis.¹ Most newly diagnosed pleural mesothelioma cannot be treated locally, and require systemic treatment with chemotherapy, but this tumor is largely chemo-resistant, and therapeutic options are limited. Indeed, consistent data are only available on first-line treatment, and options after progression are currently unclear.^{1,4} Despite this being a highly aggressive tumor, dissemination is generally local, and symptoms caused by compression and direct invasion cause the patient to deteriorate rapidly. Distant dissemination, as observed in our patient, is much more uncommon, and the development of musculoskeletal metastasis despite control of the pulmonary disease is exceptional. These events suggest that long-term tumors might disseminate atypically to the so-called "sanctuary organs", where the tumor can evade oncological control. More food for thought is that in oligometastatic patients, continuing systemic treatment while adding a local treatment appears to be a valid therapeutic option.

Our case illustrates a pleural mesothelioma with an atypical course: unusual features include the patient's long survival on oncological treatment (36 months, when mean survival is around 12 months), and an exceptionally rare site of dissemination.

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Bronchobiliary Fistula, a Late Complication of Liver Surgery $\dot{}^{\dot{}}$



Fístula biliobronquial, una complicación tardía de la cirugía hevática

To the Editor,

Bronchobiliary fistula is an abnormal communication between the bile duct and the bronchial tree. In 1850, Peacock described the first case in a 20-year-old woman with hepatic echinococcosis. It is a rare entity, and few cases have been reported in the literature. Morbidity and mortality are high, and the diagnosis is essentially clinical, guided by the pathognomonic presence of biliptysis, or bile in the sputum. Bile has a corrosive effect on the lung and pleural space.

We report the case of a 57-year-old man who presented in the emergency room with a 3-day history of fever 38.5 °C and bilous expectoration. Significant history included sigma

adenocarcinoma with liver metastases, for which he received neoadjuvant chemotherapy with irinotecan and cetuximab. Portal vein embolization and surgical resection of the liver metastasis (right hepatectomy, section of segment 4 A, and left lobe), cholecystectomy and splenectomy were subsequently performed. He received single-agent cetuximab until sigmoidectomy with colorectal anastomosis, which was performed 8 months before he presented in the emergency room. At that time, the sigma cancer was in remission (CEA 3.9 ng/ml), and the patient was being treated with cetuximab every 15 days. On physical examination, arterial blood pressure was 130/80 mmHg, heart rate 115 bpm, temperature 37.5 °C, and oxygen saturation 89%. Poor general condition, conjunctival icterus, reduced breath sounds in the right field with crackles, and hepatomegaly of 2 finger breadths. Clinical laboratory tests showed significant leukocytosis (25 790/µl) with neutrophilia (80%). Chest X-ray revealed consolidation in the right lower lobe. Given the likelihood of a biliopulmonary fistula as a late complication of liver surgery, a computed tomography (CT) of the chest-abdomen was requested, which showed parenchymal

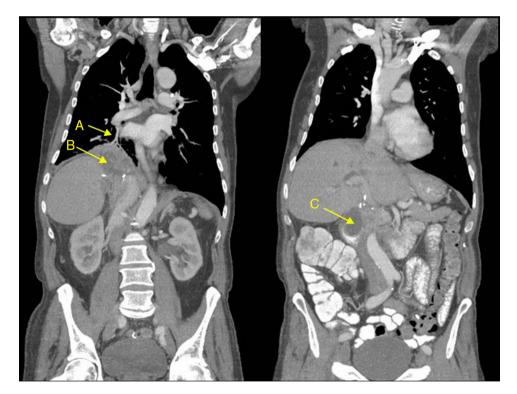


Fig. 1. Multiplanar coronal CT reconstruction slices with intravenous contrast medium. (A) Fistulous tract between the biliary tree and the bronchus of the right lower lobe. (B) Right subphrenic collection. (C) Subhepatic collection.

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