Spontaneous Hemotherax as a Presenting Form of Bronchogenic Carcinoma

**Hemotórax espontáneo como una forma de presentación del carcinoma broncogénico**

To the Editor,

Spontaneous hemotherax is a very rare form of presentation of bronchogenic carcinoma. We present the case of a 73-year-old woman, non-smoker, with a history of hypertension, dyslipidaemia, diabetes mellitus, psoriasis and depressive disorder. The patient attended the emergency department with a one-week history of pleuritic chest pain and two-month history of weight loss and asthenia. Physical examination revealed absence of breath sounds in the base of the left hemithorax. The chest radiograph showed a radiopaque image in the left base consistent with pleural effusion and contralateral mediastinal shift. Computed tomography (CT) was performed, in which loculated pleural effusion, atelectasia of the lower lobe and lingula, and a solitary pulmonary nodule (maximum 2 cm in diameter) were observed in the apical segment of the right lower lobe (Fig. 1). Thoracentesis was subsequently carried out, in which hemotherax was diagnosed and treated with chest drainage. The follow-up radiograph showed expansion, so the drain was removed on the sixth day. A new CT scan was then performed in which complete resolution of the hemotherax was observed, with persistence of the previously described nodule. The presence of malignant cells suggestive of adenocarcinoma was observed in 2 pleural fluid cytology specimens. Immunohistochemical analyses were positive for MOC-31, and a polymerase chain reaction (PCR) study showed a mutation in exon 19 of the EGFR gene. The patient was discharged with a diagnosis of EGFR-positive large cell carcinoma and treated with gefitinib.

Spontaneous hemotherax is a rare entity and its causes include neoplasms, anticoagulant treatment or coagulation disorders, endometriosis, pulmonary infarction and pneumothorax with adhesions. Infectious causes have also been described. From a neoplastic perspective, metastases from gynaecological tumours, choriocarcinoma and sarcoma are the most common causes. The primary carcinoma most often associated with the development of spontaneous hemotherax is usually schwannoma of Von Recklinghausen disease and angiosarcoma. Spontaneous hemotherax as a form of presentation of a bronchogenic carcinoma is unusual in the literature, even in the context of pleural metastases. Compression and ischaemic necrosis of adjacent lung tissue due to subpleural growth of the tumour, or the invasion of pulmonary vessels with rupture and drainage into the pleural cavity, have been suggested as possible mechanisms. We performed a literature search in this respect. In databases such as PubMed (www.ncbi.nlm.nih.gov/PubMed), we found 576 studies using the key words “spontaneous hemotherax”. Of these, only 2 were related with bronchogenic carcinoma, one described by Chou et al. due to an adenocarcinoma with ipsilateral mediastinal node metastases, and another by Ausín et al. in 2005 due to a large cell carcinoma with liver and bone metastases. In our case, pathological, immunohistochemical and biological studies were carried out, so the patient was discharged with specific treatment to which she responded well.

References


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Mediastinal Castleman’s Disease: Case Report

**Enfermedad de Castleman mediastínica: presentación de un caso**

To the Editor,

Although Castleman’s disease is a pathological entity that has been well described in the scientific literature, there are few reported cases in Spain, and they may cause problems in the differential diagnosis. We present the case of a 33-year-old woman referred to our department by Internal Medicine with a diagnosis of a mediastinal mass found on a chest radiograph performed after an accident.

The patient reported an episode of self-limiting haemoptysis one month previously, weight loss that she related with anxiety, occasional retrosternal oppression accompanied by paraesthesia in the left arm and episodes of bronchospasm. Laboratory tests were normal. The chest computed tomography (Fig. 1) revealed a soft tissue mass in the anterior mediastinum, 5 cm in diameter, occupying the prevascular space and aortopulmonary window. The lesion first suggested thymoma or lymphoma.

As a surgical approach, we chose a left anterolateral mini-thoracotomy for various reasons: it provides good access to the lesion for biopsy, but at the same time can be easily widened if the lesion is resectable and, moreover, a sub-mammary incision is more cosmetic than an anterior mediastinotomy. A rounded, well-defined mass was found in the mediastinum, which abutted the superior pulmonary vein and artery, pericardium and left upper lobe, but did not infiltrate any of these structures and was completely excised.

The specimen was sent as an intraoperative biopsy and diagnosed as thymoma.

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Definitive pathology: 6.16 x 4.67 cm rounded, well-defined tumour. Benign giant lymph node hyperplasia consistent with Castleman’s disease, histological sub-
type hyaline-vascular.

Castleman’s disease (also known as angiofollicular lymph node hyperplasia or
benign giant lymph node hyperplasia), first described by Benjamin Castleman in
1956, is a rare entity characterised by non-tumour proliferation of lymphoid tis-
tue. The cause is unknown, although a subgroup of cases is associated with human
herpesvirus-8 (HHV-8).2

It affects both sexes equally and can appear at any age and in any part of the
body, although it occurs mainly in the thorax (70%), followed by the neck (15%) and
the abdomen-pelvis (15%).

Castleman’s disease is classified into 3 histological subtypes: vascular-hyaline,
plasma cell type and mixed.3 More recently though, a new classification into 4 var-
ant has been proposed4: vascular-hyaline, plasma cell type, HHV-8-associated and
non-specific multicentric Castleman’s disease. The vascular-hyaline type is the most
common variant (90%). It usually affects young people and is associated with the
localised form. The plasma cell type represents 9% of cases, appears in adulthood
and is usually characteristic of the generalised or multicentric form.

Clinically it can present in localised or multicentric form. The localised form
appears more frequently in the thorax. The mean age is around 35 years and it affects
both sexes equally. It usually presents as an asymptomatic, isolated mass, often di-
nosed as a casual finding. It occasionally causes symptoms of compression, such
as chest or abdominal pain. Peripheral lymphadenopathies are rare and laboratory
tests show abnormalities in only 25% of cases. The multicentric form usually appears
in older patients, affects more than one area of the body and presents with sys-
temic manifestations: asthma, weight loss, fever, generalised lymphadenopathies,
skin lesions, neuropathy and hepatosplenomegaly. Laboratory tests usually show
increased ESR, anaemia and hyper gammaglobulinaemia. Unlike the localised form,
it is strongly associated with immunosuppression and HHV-8 infection.5 Its cli-
cal evolution is often aggressive and fatal due to the development of infections and
neoplasia, such as Kaposi’s sarcoma and lymphomas.

The disease is suspected using imaging techniques, but definitive diagnosis
requires a biopsy. The differential diagnosis includes other lymph node diseases,
particularly lymphomas and tuberculosis, and indeed Bonekamp et al. called it “the
great mimic.”5

Treatment in the localised form is surgical resection5; recurrence has not been
reported in most series, even with incomplete resection. The multicentric form
requires multimodal therapies with radiotherapy, chemotherapy, corticosteroids
and monoclonal antibodies.5

Conflict of Interest

The authors do not have any conflicts of interest.

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