

## Spontaneous Hemothorax as a Presenting Form of Bronchogenic Carcinoma<sup>☆</sup>

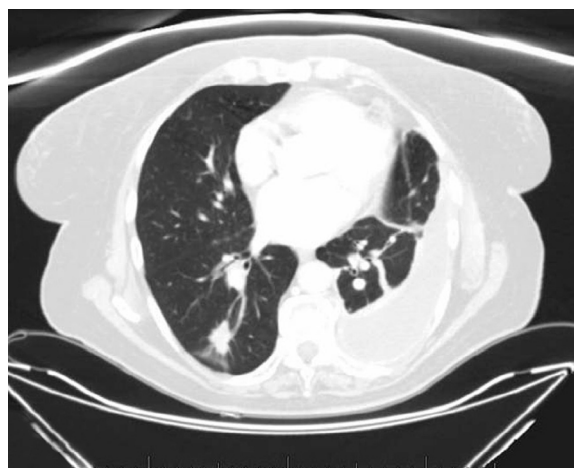
### *Hemotórax espontáneo como una forma de presentación del carcinoma broncogénico*

To the Editor,

Spontaneous hemothorax 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 hemothorax 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 hemothorax 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 hemothorax 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.<sup>1</sup> From a neoplastic perspective, metastases from gynaecological tumours, choriocarcinoma and sarcoma are the most common causes.<sup>2</sup> The primary carcinoma most often associated with the development of spontaneous hemothorax is usually schwannoma of Von Recklinghausen disease and angiosarcoma.<sup>1</sup> Spontaneous hemothorax as a form of presentation of a bronchogenic carcinoma is unusual in the

<sup>☆</sup> Please cite this article as: Ávila Martínez RJ, et al. Hemotórax espontáneo como una forma de presentación del carcinoma broncogénico. Arch Bronconeumol. 2013;49:455.



**Figure 1.** The chest CT scan shows loculated pleural effusion, atelectasia of the lower lobe and lingula, and a 2 cm solitary pulmonary nodule in the apical segment of the left lower lobe.

literature,<sup>3</sup> even in the context of pleural metastases.<sup>1</sup> 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.<sup>3</sup> We performed a literature search in this respect. In databases such as PubMed ([www.ncbi.nlm.nih.gov/PubMed](http://www.ncbi.nlm.nih.gov/PubMed)), we found 576 studies using the key words "spontaneous hemothorax". Of these, only 2 were related with bronchogenic carcinoma, one described by Chou et al.<sup>3</sup> due to an adenocarcinoma with ipsilateral mediastinal node metastases, and another by Ausín et al.<sup>2</sup> 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

1. Ali HA, Lippman M, Mundathaje U, Khaleeq G. Spontaneous hemothorax a comprehensive review. *Chest*. 2008;134:1056-65.
2. Ausín P, Gómez-Caro A, Pérez Rojo R, Moradiellos FJ, Díaz Hellín V, Martín de Nicolás JL. Spontaneous hemothorax caused by lung cancer. *Arch Bronconeumol*. 2005;41:400-1.
3. Chou SH, Cheng YJ, Kao EL, Chai CY. Spontaneous haemothorax: an unusual presentation of primary lung cancer. *Thorax*. 1993;48:1185-6.

Régulo José Ávila Martínez,<sup>a,\*</sup> Ana Hernández Voth,<sup>b</sup>  
Victoria Villena Garrido<sup>b</sup>

<sup>a</sup> Servicio de Cirugía Torácica, Hospital Universitario 12 de Octubre, Madrid, Spain

<sup>b</sup> Servicio de Neumología, Hospital Universitario 12 de Octubre, Madrid, Spain

\* Corresponding author.

E-mail address: [reguloavila@hotmail.com](mailto:reguloavila@hotmail.com)

(R.J. Ávila Martínez).

## Mediastinal Castleman's Disease: Case Report<sup>☆</sup>

### *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.

<sup>☆</sup> Please cite this article as: Miguélez Vara C, et al. Enfermedad de Castleman mediastínica: presentación de un caso. Arch Bronconeumol. 2013;49:455-6.

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.

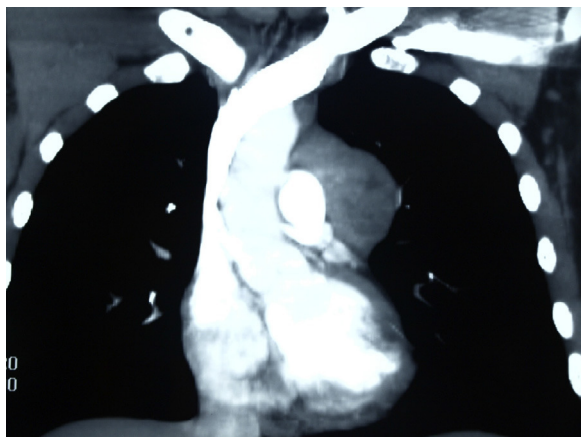


Fig. 1. Chest computed tomography.

Definitive pathology: 6.16 × 4.67 cm rounded, well-defined tumour. Benign giant lymph node hyperplasia consistent with Castleman's disease, histological subtype 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,<sup>1</sup> is a rare entity characterised by non-tumour proliferation of lymphoid tissue. The cause is unknown, although a subgroup of cases is associated with human herpesvirus-8 (HHV-8).<sup>2</sup>

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.<sup>3</sup> More recently though, a new classification into 4 variants has been proposed<sup>4</sup>: 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 diagnosed 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 systemic manifestations: asthenia, weight loss, fever, generalised lymphadenopathies, skin lesions, neuropathy and hepatosplenomegaly. Laboratory tests usually show increased ESR, anaemia and hypergammaglobulinaemia. Unlike the localised form, it is strongly associated with immunosuppression and HHV-8 infection.<sup>2</sup> Its clinical 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".<sup>5</sup>

Treatment in the localised form is surgical resection<sup>6</sup>; 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.<sup>5</sup>

## Conflict of Interest

The authors do not have any conflicts of interest.

## References

1. Castleman B, Iverson L, Menendez VP. Localized mediastinal lymph node hyperplasia resembling thymoma. *Cancer*. 1956;9:822–30.
2. Katano H, Sata T. An attractive relation of human herpesvirus-8 with multicentric Castleman's disease. *Intern Med*. 1999;38:221.
3. Keller AR, Hocholzer L, Castleman B. Hyaline-vascular and plasma-cell types of giant lymph node hyperplasia of the mediastinum and other locations. *Cancer*. 1972;29:670–83.
4. Cronin DM, Warnke RA. Castleman disease: an update on classification and the spectrum of associated lesions. *Adv Anat Pathol*. 2009;16:236–46.
5. Bonekamp D, Horton K, Hruban R, Fishman E. Castleman disease: the great mimic. *Radiographics*. 2011;31:1793–807.
6. Bowne WB, Lewis JJ, Filippa DA, Niesvizky R, Brooks AD, Burt ME, et al. The management of unicentric and multicentric Castleman's disease. *Cancer*. 1999;85:706–17.

Carlos Miguélez Vara,<sup>a,\*</sup> Manuel Mariñán Gorospe,<sup>a</sup> Irune Albistur Lesmes<sup>b</sup>

<sup>a</sup> Servicio de Cirugía Torácica, Hospital San Pedro, Logroño, Spain

<sup>b</sup> Servicio de Medicina Interna, Hospital San Pedro, Logroño, Spain

\* Corresponding author.

E-mail addresses: cmiguel@riojasalud.es, cmvara61@gmail.com (C. Miguélez Vara).