



CASE FOR DIAGNOSIS

A 75-year-old female with dyspnoea and chest pain

M. Tresinie*, F. Van De Maele[#], W. Verhaeghe*, B. Van de Maele*, R. Venken[†] and K. Hertveldt⁺

CASE HISTORY

A 75-yr-old nonsmoking female, with an unremarkable medical history, was admitted to the emergency ward for evaluation of dyspnoea and right-sided thoracic pain.

Physical examination revealed tachycardia and decreased breath sounds at the right lower base, with dullness on percussion. Electrocardiography showed atrial fibrillation.

A right-sided hemithoracic mass was seen upon chest radiography (fig. 1). Computed tomography (CT) of the chest and magnetic resonance (MR) were also performed (figs 2 and 3).

Laboratory results were as follows: white blood cells = 13.42×10^6 cells·L⁻¹; red blood cells = 451×10^6 cells·L⁻¹ and

serum total protein $66 \text{ g} \cdot \text{L}^{-1}$ (albumin = 55.1%, gammaglobulin = 12.1%). Carcinoembryonic antigen, β_2 -microglobulin, α -fetoprotein and β -human chorionic gonadotropin were all within normal range.

Abdominal ultrasound revealed a liver cyst. Bone scintigraphy and CT of the pelvic region were normal.

A right-sided posterolateral thoracotomy was performed. A 10-cm large, soft, encapsulated, white mass was found to be attached to the parietal pleura, without any signs of local infiltration or adhesions with the adjacent lung. No pedicle was found. Complete removal was easily performed. The results of the microscopical examination are shown in figures 4–6.



FIGURE 1. Chest radiograph.



FIGURE 2. Computed tomography of the chest.

*Respiratory Division, [†]Dept of Surgery, and ⁺Dept of Pathology, Henri Serruys Hospital, and [#]Torhoutsesteenweg 51, B-8400, Oostende, Belgium.

CORRESPONDENCE: B. Van de Maele, Respiratory Division, Henri Serruys Hospital, Kairostraat 84, B-8400 Oostende, Belgium. Fax: 32 59555792. E-mail: dr.vandemaele@henriserruysav.be

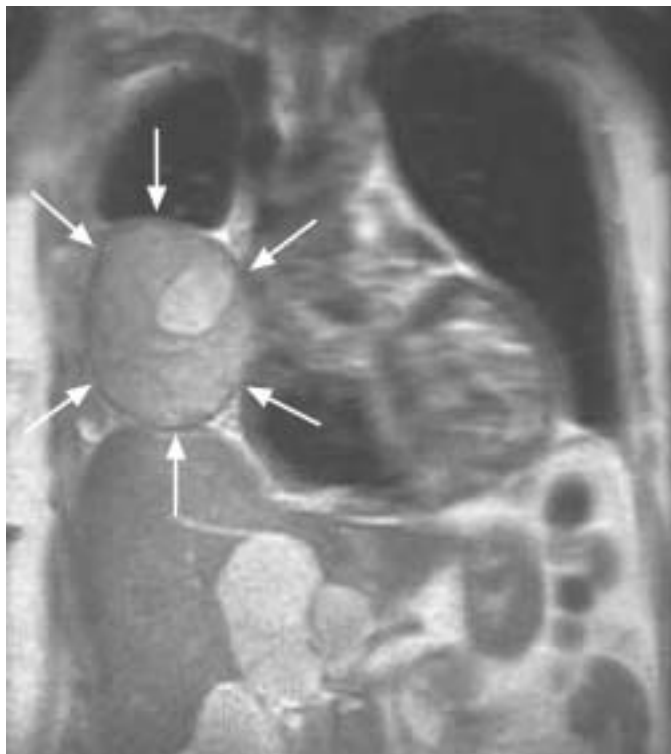


FIGURE 3. Magnetic resonance tomography of the chest and abdomen (T2-weighted image).

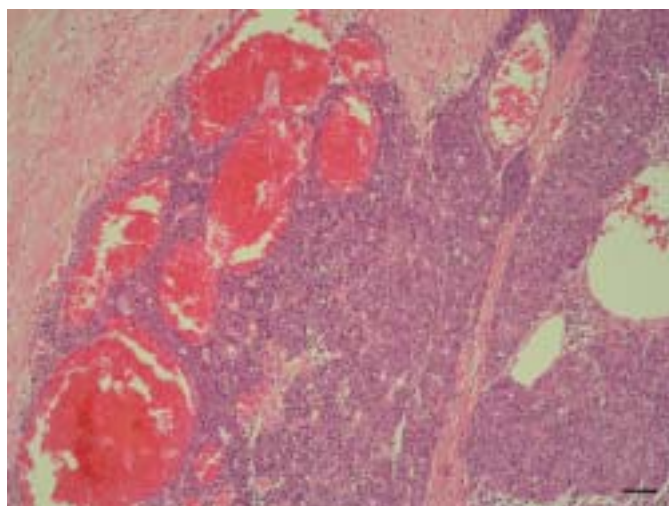


FIGURE 4. Histological picture of the open-chest biopsy. Haematoxylin-eosin stain. Scale bar=50 μ m.

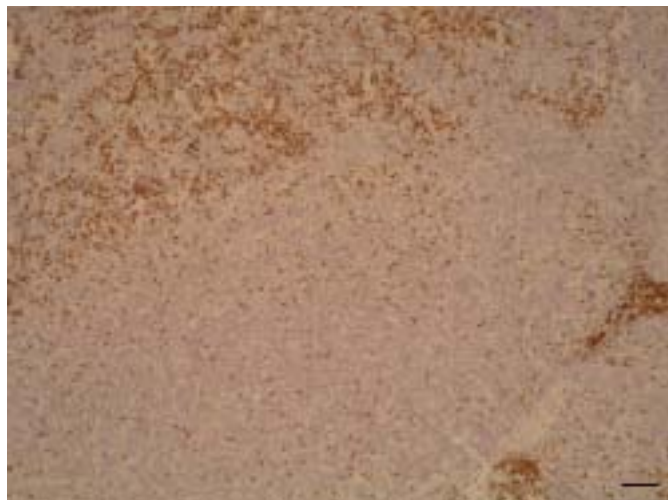


FIGURE 5. CD3 antibody immunostain. Scale bar=50 μ m.

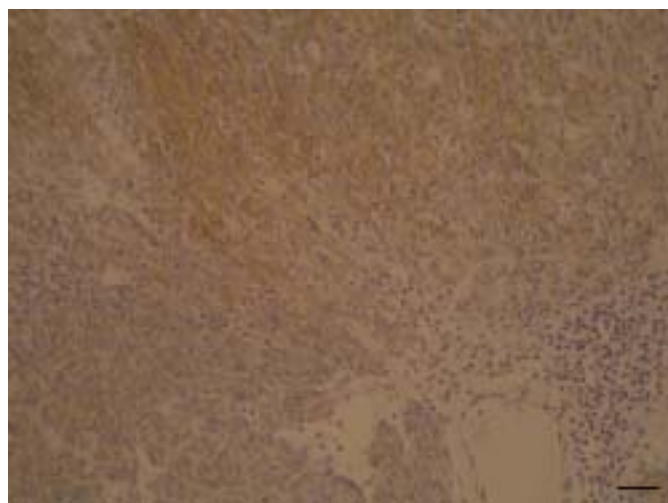


FIGURE 6. Broad spectrum cytokeratin immunostain. Scale bar=25 μ m.

BEFORE TURNING THE PAGE, INTERPRET THE RADIOLOGICAL PICTURES AND THE HISTOLOGICAL FIGURES AND SUGGEST A DIAGNOSIS.

INTERPRETATION

A right-sided hemithoracic mass with secondary compressive atelectasis of the right lower lobe was seen upon chest radiography (fig. 1).

CT of the chest showed a large tumour mass at the right lower lobe with associated pleural effusion and compressive atelectasis of the right lower lobe. No lymph nodes were seen (fig. 2).

Inhomogeneous signal intensities with scattered high-intensity areas were seen on T2-weighted MR imaging (fig. 3). T1-weighted MR imaging depicted a well encapsulated, heterogeneous, 8×7×12 cm, isointense to slightly hyperintense lesion, while Gd-enhanced T1-weighted MR images demonstrated strong tumoural enhancement.

Histopathology showed an intrapleural mass composed of bland looking spindle cells intermingled with small lymphocytes (fig. 4). The aggregates of small lymphocytes were all immature CD3-antibody positive T-lymphocytes (fig. 5). A broad spectrum cytokeratin staining was diffusely positive in the spindle cell population (fig. 6).

Diagnosis: Primary pleural thymoma.

DISCUSSION

Thymomas are defined as tumours originating in the epithelial components of the thymus. Although they are usually found in the anterior mediastinum, thymomas have also been reported in aberrant locations, such as the right submandibular region, adjacent to the thyroid gland, the trachea, pulmonary hilum or even within the pulmonary parenchyma.

Ectopic thymoma with involvement of the pleura has only been rarely reported in the English literature [1–3]. In those cases, as in the presented patient, radiographic and surgical exploration excluded pleural involvement due to direct extension or pleural dissemination from a mediastinal tumour. The latter creating a pitfall in the differential diagnosis of pleural-based lesions.

The histogenesis of primary pleural thymoma remains obscure. One possible explanation is that of embryological displacement. The thymic primordials appear late in the 6th week of gestation from a sacculation of the third pharyngeal pouch. They form two epithelial bars that fuse in the midline within the anterosuperior mediastinum. During this descent, the tail portion of the organ becomes thin and breaks up into small fragments; these may persist and give rise to ectopic thymic tissue and thymomas [4].

Thymomas can frequently be associated with a variety of paraneoplastic disorders, such as myasthenia gravis [5], pure

red cell aplasia [6] and hypogammaglobulinaemia [7]. None of these were observed in the current patient.

As a result of the paucity of studies on primary pleural thymomas and their incomplete follow-up information, the standard procedure for treatment has not been adequately delineated.

Thymomas are slowly growing tumours that may be adequately treated by surgical resection alone, when completely confined to the lung. It is still a matter of debate whether radiation therapy should be used for completely resected invasive thymomas [8]. Recent studies have indicated that whole mediastinum irradiation is effective in preventing mediastinal recurrence [8]. However, in patients with pleural invasion of the tumour determined by microscopic examination, mediastinal irradiation alone was insufficient to avoid pleural-based recurrence [9].

REFERENCES

- 1 Moran CA, Travis WD, Rosado-de-Christenson M, Koss MN, Rosai J. Thymomas presenting as pleural tumors: report of eight cases. *Am J Surg Pathol* 1992; 16: 138–144.
- 2 Moran CA, Suster S, Fishback NF, Koss MN. Primary intrapulmonary thymoma: a clinopathological and immunohistochemical study of eight cases. *Am J Surg Pathol* 1995; 19: 304–312.
- 3 Shih DF, Wang JS, Tseng HH, Tiao WM. Primary pleural thymoma. *Arch Pathol Lab Med* 1997; 121: 79–82.
- 4 Kadouch R, Thoreux PH, Lucas C, Pencole-Nouveau C, Tas P, Kernec J. Pleural thymoma. Apropos of a case and review of the literature. *J Radiol* 1989; 70: 491–496.
- 5 Siao P, Zulkerberg LR. Case 15-2000: A 69 year-old man with myasthenia gravis and a mediastinal mass. *N Engl J Med* 2000; 342: 1508–1514.
- 6 Masaoka A, Hashimoto T, Shibata K, Yamakawa Y, Nakamae K, Iizuka M. Thymomas associated with pure red cell aplasia. Histologic and follow-up studies. *Cancer* 1989; 64: 1872–1878.
- 7 Oshiriki T, Morikawa T, Sugiura H, Katoh H. Thymoma associated with hypogammaglobulinemia (Good's syndrome): report of a case. *Surg Today* 2002; 32: 264–266.
- 8 Graeber GM, Tamim W. Current status of the diagnosis and treatment of thymoma. *Semin Thorac Cardiovasc Surg* 2000; 12: 268–277.
- 9 Kazuhiko O, Takashi U, Takafumi T, et al. Postoperative radiotherapy for patients with completely resected thymomas: a multi-institutional, retrospective review of 103 patients. *Cancer* 2000; 94: 1405–1413.