CASE REPORTS

Localized Fibrous Tumors of the Pleura: Clinical and Surgical Evaluation

J.M. Galbis Caravajal,[a] J.G. Sales Badía,[a] M. Navarro Hervás,[b] J.J. Mafé Madueño,[c] P. Cordero Rodríguez,[d] and J.M. Rodríguez Paniagua[c]

[a]Servicio de Cirugía Torácica, Hospital de La Ribera, Alzira, Valencia, Spain.
[b]Servicio de Anatomía Patológica, Hospital de La Ribera, Alzira, Valencia, Spain.
[c]Servicio de Cirugía Torácica, Hospital General Universitario de Alicante, Alicante, Spain.
[d]Servicio de Neumología, Hospital de La Ribera, Alzira, Valencia, Spain.

Solitary fibrous tumors of the pleura are uncommon and mainly arise in the pleura itself. Such tumors are generally asymptomatic and slow-growing. We report a series of 10 cases (8 men and 2 women with a mean age of 58.6 years) treated over a period of 54 months. The tumors were classified histologically as benign or malignant according to the criteria used by England. The treatment of choice was complete resection of the tumor.

Six posterolateral thoracotomies and 4 video-assisted resections were performed. Histology showed a mixture of fibroblast-like cells and collagenous stroma. Sarcomatous degeneration was observed in the excised tumor of 1 patient. The patients were followed for a mean of 23.9 months.

We conclude that although fibrous tumors of the pleura are considered benign histologically, complete resection and follow up for all patients are recommended.

Key words: *Fibrous tumor of the pleura. Pleura. Mesothelioma.*

Tumores fibrosos de localización pleural: valoración clinicoquirúrgica

Los tumores fibrosos pleurales solitarios son lesiones infrecuentes que en su mayoría derivan de la pleura. Generalmente asintomáticos, poseen un crecimiento lento. En el presente trabajo se describe una serie de 10 casos (8 varones y 2 mujeres, con una edad media de 58,6 años) tratados en un período de 54 meses. Histológicamente se clasificaron como benignos o malignos basándose en los criterios de England. El tratamiento de elección fue la cirugía con criterios de resección completa.

Se realizaron 6 toracotomías posterolaterales y 4 resecciones por videocirugía. Microscópicamente estaban constituidos por células de aspecto fibroblástico, entremezcladas con estroma colagenizada. Uno de los pacientes presentó una degeneración sarcomatosa en la pieza. Se siguió a los pacientes en consultas, con un seguimiento medio de 23,9 meses.

En conclusión, los tumores fibrosos pleurales, aunque considerados histológicamente benignos, precisan de la resección completa. Se recomienda el seguimiento evolutivo en todos los pacientes.

Palabras clave: *Tumor fibroso pleural. Pleura. Mesotelioma.*

Introduction

Solitary fibrous tumors of the pleura are not common. Most arise in the pleura itself and only 7.5% are intraparenchymatous. Such tumors are generally asymptomatic with slow intrathoracic growth leading to the compression of adjacent structures. Preliminary diagnosis is based on radiological examination and confirmed through histological study of samples obtained by fine needle aspiration or through analysis of the surgical specimen. Resection of the tumor is usually associated with good outcome and has a low index of recurrence if the resection is complete and free of microscopic involvement.

Just over 800 cases of such tumors have been described in the literature.[1] In the last 2 decades the mesenchymal origin of solitary fibrous tumors of the pleura has been established through immunohistochemical studies indicating they are negative for cytokeratin expression and positive for CD34.

We report a series of 10 cases treated in 2 hospitals over a period of 54 months. We discuss the clinical presentation, surgical treatment, and follow up.

Case Description

The 10 patients whose cases are reviewed (Table 1) were diagnosed and treated in our 2 hospitals. Sex distribution was 8 men and 2 women with a mean age of 58.6 years (range,

Correspondence: Dr. J.M. Galbis.
C/ Ayora 35, 1.º, 1.ª. 46018 Valencia. España.
E-mail: jgalbis@hospital-ribera.com

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TABLE 1
Patient Characteristics*

Case	Sex	Age, Years	Location	Symptoms	FNA	Surgery	Size, cm	Follow-up, Months	Survival
1	M	68	PPL	No	Yes	THOR	6	54	Yes
2	F	73	PPL	No	Yes	THOR	7	35	Yes
3	F	70	VPL	No	No	VAT	8	26	Yes
4	M	33	VPL	No	No	VAT	5	18	Yes
5	M	35	VPL	No	No	VAT	7	23	Yes
6	M	38	VPL	No	Yes	VAT	4	7	Yes
7	M	76	PPL	Yes	Yes	THOR	15	6	No
8	M	75	PAREN	Yes	Yes	THOR	15	37	Yes
9	M	50	VPL	No	Yes	THOR	8	15	Yes
10	M	68	PPL	No	Yes	THOR	8	18	Yes

*M indicates male; F, female; VPL, visceral pleura; PPL, parietal pleura; PAREN, intraparenchymatous; FNA, fine needle aspiration; THOR, thoracotomy, and VAT, video-assisted thoracoscopy.

33-76 years). Two patients were diagnosed based on clinical presentation: nonspecific pleuritic pain in 1 patient and progressive increase of dyspnea in the other. Ipsilateral pleural effusion coincided with the tumor in 1 patient. Radiographs and computed tomography scans of the chest as well as bronchoscopy were performed on all patients. Fine needle aspiration was performed in 7 patients but gave inconclusive results. All patients underwent resection, and tumors were histologically classified as benign or malignant based on the criteria of England et al[2] (Table 2).

TABLE 2
Histological Criteria of Malignancy of England et al[2]

>4 mitotic figures ×10 high-power fields
Hemorrhage
High cellularity
Pleomorphism
Necrosis

Results

Six posterolateral thoracotomies and 4 video-assisted thoracoscopies were performed. Nine atypical resections were carried out as well as a right lower lobectomy when the location was intraparenchymal. All resections were complete, and subsequent histological examinations revealed the surgical margins to be negative.

To the naked eye tumors appeared round or ovoid in shape. They were encapsulated and attached to the visceral, parietal, or, in 1 case, the intraparenchymal pleura by a pedicle. Tumor size ranged between 4 and 15 cm. Under the microscope (Figure 1), the tumors were seen to be composed of fibroblast-like cells embedded in a collagenous stroma and were immunohistochemically positive for CD34. The definitive histological diagnosis was benign fibrous tumor of the pleura in all cases, and in 1 case (with a 15-cm tumor) the biopsy showed sarcomatous transformation (resulting in death 6 hours after surgery due to cardiogenic shock with fulminant pulmonary edema). Patients were followed up for a mean 23.9 months (range, 6-54 months). No signs of recurrence in any of the patients have been reported.

Discussion

Fibrous tumors of the pleura account for less than 5% of pleural tumors and are mostly asymptomatic. We detected only 2 cases upon clinical observation, and in both the tumors were large in size (15 cm).

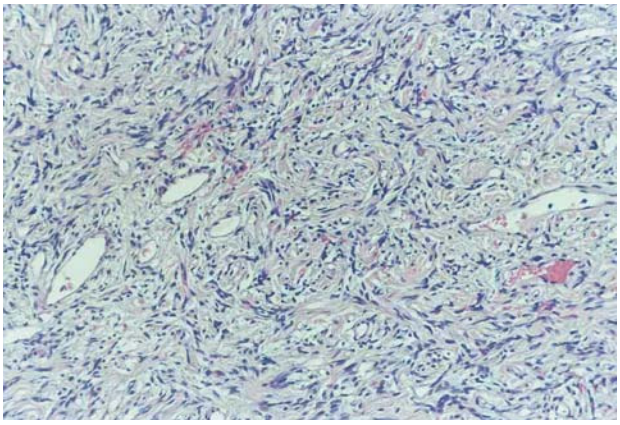

Figure 1. Hematoxylin-eosin. Spindle cells enmeshed in collagenous stroma.

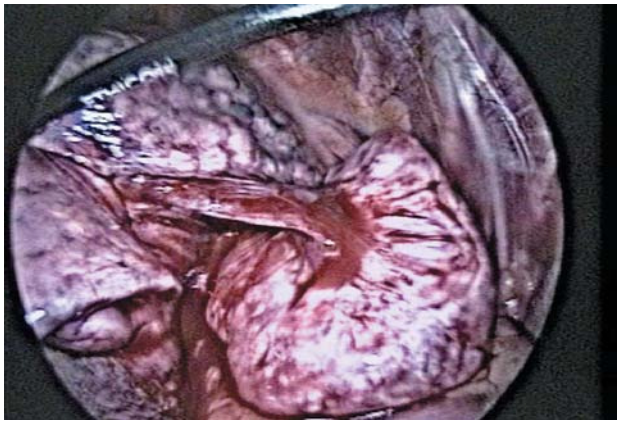

Figure 2. Video-assisted thoracoscopic image of a fibrous tumor attached to the visceral pleura (case 4).

Extrathoracic signs and symptoms include hypoglycemia,[3] galactorrhea, weight loss, arthritic pain, and hypertrophic osteoarthropathy.[4] One patient in our series had associated hypertrophic osteoarthropathy. Twenty percent of the cases in the literature[5] report such an association, which is related to the increased production of hyaluronic acid by tumor cells. Radiographic studies (x-rays and computed tomography scans of the chest) are the diagnostic tests of choice.[6] Some authors emphasize the diagnostic role of magnetic resonance imaging in differentiating the fibrous nature of the lesion.[7]

We performed fine needle aspiration in 7 patients, and although the results were inconclusive in all cases, the technique provided grounds for suspicion in 1 case. One series in the literature reported needle aspiration diagnosis as having a low diagnostic yield.[8]

The treatment of choice is complete resection.[5,9] We performed thoracotomies in 6 patients, and video-assisted thoracoscopies in 4 (Figure 2). The choice of technique was chosen based on size of the tumor and detection of a pedicle, which would facilitate video-assisted resection (with tissue removal through a lengthened access incision). When the parietal pleura is involved, extrapleural resection, including the adjacent chest wall, is advised.[10]

The best predictor of a good prognosis is complete resection.[11,12] In our series we have observed neither recurrence of the disease nor distant metastasis in any of the patients. England et al[2] reported recurrence in only 1.4% of their patients (out of a total of 141 cases classified as benign tumors). Cardillo et al[5] saw recurrence in only 1 patient, whose tumor was classified as malignant, 13 months after surgery.

Adjuvant treatments with chemo- and radiotherapy have been used although the benefits have not yet been established.[13]

In conclusion, solitary fibrous tumors of the pleura are confined to the pleura and are mainly asymptomatic. While considered benign histologically, complete resection is the treatment of choice. Follow up of all patients is advisable.

REFERENCES

1. de Perrot M, Fischer S, Bründler MA, Sekine Y, Keshavjee S. Solitary fibrous tumors of the pleura. Ann Thorac Surg 2002; 74:28593.
2. England DM, Hocchholzer L, McCarty MJ. Localized benign and malignant fibrous tumor of the pleura. A review of 223 cases. Am J Surg Pathol 1989;13:640-58.
3. Luque Sánchez F, Ruiz Zafra J, Sánchez Palencia A. Hipoglucemia grave secundaria a un mesotelioma fibroso pleural localizado. Arch Bronconeumol 1999;35:102-3.
4. Altinok T, Topçu S, Tastepe A, Yazici U, Çetin G. Localized fibrous tumors of the pleura: clinical and surgical evaluation. Ann Thorac Surg 2003;76:892-5.
5. Cardillo G, Facciolo F, Cavazzana A, Capece G, Gasparri R, Martelli M. Localized (solitary) fibrous tumors of the pleura: an analysis of 55 patients. Ann Thorac Surg 2000;70:1808-12.
6. Versluis PJ, Lamers RJ. Localized pleural fibroma: radiological features. Eur J Radiol 1994;18:124-5.
7. de Perrot M. Fibrous tumors of the pleura. Current Treatment Options in Oncology 2000;1:293-8.
8. Ali SZ, Hoon V, Hoda S, Heelan R, Zakowski M. Solitary fibrous tumor. Cancer 1997;81;116-21.
9. Rena O, Filosso Pl, Papalina E, Molinatti M, Di Marzio P, Oliaro A. Solitary fibrous tumour of the pleura: surgical treatment. Eur J Cardiothorac Surg 2001;19:185-9.
10. de Perrot M, Kurt AM, Robert JH, Borisch B, Spiliopoulos A. Clinical behavior of solitary fibrous tumors of the pleura. Ann Thorac Surg 1999;67:1456-9.
11. Blanco M, Montero C, Rivas J, Otero I, Yebra MT, Verea H. Mesotelioma fibroso benigno: aportación de 8 casos. Arch Bronconeumol 1998;34:361-3.
12. Magdeleinat P, Alifano M, Petino A, le Rochais JP, Dulmet E, Galateau F, et al. Solitary fibrous tumors of the pleura: clinical characteristics, surgical treatment and outcome. Eur J Cardiothorac Surg 2002;21:1087-93.
13. Suter M, Gebhard S, Boumghar M, Peloponisios N, Genton CY. Localized fibrous tumors of the pleura: 15 new cases and review of the literature. Eur J Cardiothorac Surg 1998;14:453-9.