• Pleural solitary fibrous tumors (SFTs) are uncommon tumors. Although these tumors have been well characterized, malignant pleural SFTs with liposarcomatous differentiation have not been reported. We report an unusual malignant pleural SFT intermixed with well-differentiated liposarcoma. The patient was a 66-year-old, white man with a large, solid right pleural mass that measured 13.5 × 10.3 × 8.5 cm. The tumor was composed of spindle-shaped and plump cells embedded in dense collagenous stroma. The tumor cells were arranged in interlacing fascicles or in a patternless pattern. Marked nuclear atypia, a high mitotic rate (21 mitoses per 10 high-power fields), and areas of prominent necrosis were evident. In addition, numerous adipocytes mixed with typical lipoblasts were seen scattered throughout portions of the tumor. Immunohistochemistry revealed the tumor cells were strongly positive for CD34 and vimentin and negative for cytokeratin, desmin, smooth muscle actin (IA4), and S100. To the best of our knowledge, this case represents the first example of a malignant SFT with liposarcomatous differentiation.

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Solitary fibrous tumors (SFTs) are uncommon tumors, which occur at both intrathoracic and extrathoracic locations.1-3 These tumors mainly affect older persons and are slightly more common in women than in men. Although most of these tumors are benign, some have developed local recurrence and metastasis.1,3 These tumors have been well characterized by hypercellularity, nuclear atypia, and a high mitotic rate and necrosis. However, nuclear pleomorphism and a high mitotic rate do not necessarily indicate a poor prognosis.1,3 Benign SFTs of the pleura require complete local resection, and the malignant tumors warrant more radical therapy.

Although malignant SFTs are well recognized histologically, to our knowledge, cases with liposarcomatous differentiation have not been reported. We describe herein an unusual malignant SFT of the pleura intermixed with well-differentiated liposarcoma.

MATERIALS AND METHODS

Routine hematoxylin-eosin-stained sections for light microscopy were examined from formalin-fixed and paraffin-embedded tissue. Immunohistochemical stains were performed on formalin-fixed and paraffin-embedded tissue using an avidin-biotin immunoperoxidase and strepavidin horseradish peroxidase. The antibodies used included CD34 (1:2, BioGenex, San Ramon, Calif), vimentin (1:2000, BioGenex), cytokeratin (AE1/AE3, 1:2000, Boehringer-Mannheim, Indianapolis, Ind), desmin (1:100, Accurate Chemical and Scientific Corp, Westbury, NY), smooth muscle actin (LA4, 1:4000, Sigma Diagnostic, St Louis, Mo), and S100 protein (1:1500, Dako Corporation, Carpenteria, Calif). Appropriate positive and negative controls were set up at the same time.

REPORT OF A CASE

The patient was a 66-year-old man who presented to Rhode Island Hospital with a large right pleural mass in February 1999. He was found to have a mass at the right lower aspect of the pleura 22 years previously when he had a chest x-ray examination following presentation with acute heartburn. This mass was thought to be a benign lesion and was followed annually. It grew slowly on subsequent computed tomography (CT) scans. In the past few years, however, it has progressively enlarged. Chest CT scans at the time he presented to Rhode Island Hospital revealed a large pleural-based mass of the right lower lobe of the lung (Figure 1). Two fine needle aspirations from the mass were inconclusive for a diagnosis due to necrotic tissue. The patient complained of an intermittent nonproductive cough. He denied any shortness of breath, pleuritic chest pain, abdominal pain, or weight loss.

His medical history was remarkable for a 30-year history of cigarette smoking, stable coronary artery disease after a myocardial infarction in 1989, and carcinoma of colon, status post resection in 1990. There was no history of tuberculosis or asbestos exposure. He previously had a part-time job working with chemicals and was, therefore, exposed to uncertain types of fumes.

The patient then underwent a right thoracotomy and a right lower lobe lobectomy. A large multilobulated bulky mass, which

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MACROSCOPIC AND MICROSCOPIC FINDINGS

The surgically resected specimen consisted of an entire right lower lobe of lung. A pleural-based tumor was found at the superior and medial basal segment of the right lower lobe. The tumor measured 13.5 × 10.3 × 8.5 cm. It was well circumscribed, and the external surface appeared intact and glistening. The cut surface was gray, tan to yellow, and predominantly solid with a slightly whorled and nodular appearance. The solid area had small areas with traversing gray-yellow bands throughout the center of the specimen. The tumor involved the pleural surface and appeared to arise from this area with a small amount of adjacent, compressed, dense, tan, brown pulmonary parenchyma noted especially in the inferior portion of the tumor. The remaining lung tissue was soft, tan, and gray, with a small amount of anthracosis without any additional mass lesions.

Microscopically, the tumor exhibited a classic histologic appearance of an SFT (Figure 2, A through D). It was composed of spindle and plump-shaped cells embedded in a dense collagenous stroma. These cells were loosely arranged in interlacing fascicles or had a short storiform pattern. Vascular clefts and branching, which mimicked hemangiopericytoma, were seen. Most of the tumor showed marked nuclear atypia and a high mitotic rate (21 mitoses per 10 high-power fields). Approximately 40% of the tumor showed prominent necrosis. In addition, numerous adipocytes mixed with typical lipoblasts were seen scattered throughout portions of the tumor. This liposarcomatous component was estimated to be approximately 15% of the total tumor. The needle biopsy specimen of the recurrence revealed foci of the malignant SFT with no obvious liposarcomatous differentiation.

Immunohistochemistry revealed the tumor cells from the malignant SFT area were uniformly and strongly positive for CD34 (Figure 3) and vimentin. They were negative for cytokeratin, desmin, smooth muscle actin (IA4), and S100. In addition, scattered spindle cells from the liposarcomatous areas were also positive for CD34 and vimentin. The lipoblasts were negative for CD34 and S100.

COMMENT

In this report, we described an unusual case of a high-grade malignant SFT of the pleura with liposarcomatous differentiation. Although liposarcomatous differentiation has been reported in diffuse mesothelioma of the pleura, it has not been previously reported in SFT of the pleura.

In the present case, the tumor was composed of 2 entirely different tumor cell populations, malignant SFT and well-differentiated liposarcoma. Although the possibility of 2 coexisting primary malignant pleural tumors cannot be totally excluded, several features suggest that this is a single tumor. The tumor described in this report was a single mass lesion and not 2 adjacent abutting tumors and was mainly composed of spindle-shaped cells embedded in a dense collagenous stroma. Liposarcomatous areas were seen scattered throughout portions of the tumor and compromised relatively small portions (approximately 15%) of the tumor mass. Primary liposarcomas of the pleura, although reported, are extremely rare. These tumors usually present as typical liposarcomas, such as well-differentiated or myxoid, which are the most common types reported. An interlacing fascicular growth pattern or a patternless pattern of the tumor cells in a dense collagenous stroma are not the characteristics of these liposarcomas. Recently, a spindle cell variant of well-differentiated liposarcoma has been reported. These tumors share certain histomorphologic and immunohistochemical features with our case. These tumors, however, like other types of liposarcomas, were located in the extremities, shoulder, and upper back and arose predominantly in subcutaneous tissue. In addition, unlike the present case, these tumors frequently have myxoid change and a better-organized fascicular pattern. Although immunoreactivity for CD34 is found in lipomatous tumors, including the recently described spindle cell variant of liposarcoma, only focal or scattered cells are positive for CD34. The uniform and strong positivity of tumor cells for CD34 in our case supports the diagnosis of malignant SFT of the pleura with liposarcomatous differentiation.

In the present case, the tumor cells are composed of spindle and plump-shaped cells, which are arranged in interlacing fascicles or a short storiform pattern. Focally, vascular clefts and branching are appreciated. These morphological features can mimic other benign or malignant lesions, such as malignant peripheral nerve sheath tumor (MPNST), malignant mesothelioma, sarcomatoid type, monophasic synovial sarcoma, malignant fibrous histiocytoma, fibrosarcoma, leiomyosarcoma, and spindle cell carcinoma. In MPNST, the cells are usually arranged in sweeping fascicles. Densely hypercellular areas alternate with hypocellular myxoid areas. The cells may be arranged in a nodular or whorled pattern. One of the important features of the MPNST is the proliferation of the tumor cells in the subendothelial zones of the blood ves-
The tumor was composed of spindle and plump cells embedded in a dense collagenous stroma (A). These cells were arranged in interlacing fascicles or had a short storiform pattern. (Inset, high-power detail of the spindle-shaped tumor cells.) Note areas with plump tumor cells, demonstrating marked nuclear atypia and numerous mitoses (arrows) (B). Adipocytes were mixed with lipoblasts in the areas of well-differentiated liposarcoma (C and D) (hematoxylin-eosin, original magnifications ×100 [A], ×200 [inset], ×400 [B], ×100 [C], and ×200 [D]).

Figure 3. CD34 immunoreactivity of the tumor cells (original magnification ×200).

Figure 2. The tumor was composed of spindle and plump cells embedded in a dense collagenous stroma (A). These cells were arranged in interlacing fascicles or had a short storiform pattern. (Inset, high-power detail of the spindle-shaped tumor cells.) Note areas with plump tumor cells, demonstrating marked nuclear atypia and numerous mitoses (arrows) (B). Adipocytes were mixed with lipoblasts in the areas of well-differentiated liposarcoma (C and D) (hematoxylin-eosin, original magnifications ×100 [A], ×200 [inset], ×400 [B], ×100 [C], and ×200 [D]).

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from benign or low-grade tumors. A case of malignant recurrence of an SFT of the pleura developing 20 years after initial surgery has been reported. In the present case, the patient had the lesion for 22 years with recent rapid growth. This could represent a transformation from a benign pleural tumor to a more aggressive malignant tumor.

To the best of our knowledge, this is the first report of a malignant SFT of the pleura with liposarcomatosus differentiation. Recognizing this heterogeneity of SFT of the pleura will be helpful in the accurate diagnosis and appropriate management of these patients. This also raises interesting suggestions about the relation between lipomatous tumors and SFT in general, and possibly the cell of origin, especially in light of the recent report on the reactivity of CD34 in lipomatous tumors. This should be addressed in future studies.

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