Primary intrapulmonary solitary fibrous tumours

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Abstract. Due to the extreme rarity of primary intrapulmonary solitary fibrous tumours (SFTs), their clinical course, imaging characteristics, diagnosis, treatment and prognosis are poorly understood. The present study therefore assessed the diagnosis and management of primary intrapulmonary SFTs and systematically reviewed previously reported cases in the literature. A total of 5 patients who underwent resection for primary intrapulmonary SFTs were enrolled in the present study and their clinical course, tumour characteristics, management and survival were assessed in this retrospective study. Relevant studies regarding primary intrapulmonary SFTs were searched using PubMed and tumour characteristics, clinicopathologic features, therapeutic strategy and survival outcomes were reviewed. Of the 5 cases, all were males, with a mean age of 57.6 years (range, 37-68 years). All patients were asymptomatic and were identified incidentally on routine computed tomography examination. A total of 3 patients underwent thoracotomy and 2 patients underwent video-assisted thoracoscopic surgery. All tumours were completely resected. Postoperative haemorrhage occurred in 1 patient and he received surgical intervention for haemostasis. The average hospital stay was 15 (4-22) days, and no mortality occurred. The mean length of the postoperative follow-up was 37.6 (1-67) months. One patient was lost to follow-up, and 4 patients were asymptomatic. A total of 19 studies were identified from database searches. They included a total of 45 patients: Twenty-three males and 22 females (mean age, 59.4 years; range, 7-81 years). A total of 12 patients were asymptomatic, and pain and coughing were the major symptoms. Five, one, two, four, and 17 tumours occurred in the right upper lobe, right middle lobe, right lower lobe, left upper lobe and left lower lobe, respectively. A total of 39 patients underwent surgery, 1 patient underwent radiotherapy, and 1 patient underwent radiofrequency ablation. A total of 22 patients were followed up and the mean length of the postoperative follow-up was 48 (1-168) months. One patient was diagnosed with chest wall metastases, and 5 patients succumbed to mortality. To conclude, primary intrapulmonary SFTs are extremely rare and typically identified incidentally. The present findings indicated that the left lower lobe was the most common site location and complete surgical resection is a safe and effective treatment.

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Introduction

A solitary fibrous tumour (SFT) is a rare, slow-growing, mesenchymal neoplasm arising from the pleura, which is unrelated to asbestos exposure or cigarette smoking (1) and was initially described in 1931 (2). Over the past 80 years, SFTs have been identified in numerous extrapleural locations, including the nasal cavity (3), breast (4), stomach (5), bronchus (6), head and neck (7), liver (8), oesophagus (9), pelvic (10), pancreas (11), prostate (12), orbit (13), central nervous system (14), parotid gland (15), kidney (16), lung (17), sella turcica (18), heart (19), conus medullaris (20), omentum (21), infratemporal fossa (22), bladder (23), soft tissues of the extremities (24), palatine tonsil (25), diaphragm (26), mesentery (27), lumbar spine (28), thymus (29), oral cavity (30), spermatic cord (31), thyroid (32), rectum (33), salivary glands (34), retroperitoneum (35), larynx (36), trachea (37), adrenal gland (38), female genital tract (39), periosteum of bone (40), mediastinum (41) and hypopharynx (42).

To our knowledge, SFTs are extremely rare in the lung (43). There are few detailed case reports concerning the clinical course, imaging characteristics, diagnosis, treatment and prognosis of primary intrapulmonary SFTs. The main purpose of the present study was to report our experience with the diagnosis and management of primary intrapulmonary SFTs and to systematically review previously reported cases in the literature.

Patients and methods

We retrospectively reviewed the records of 5 patients with primary intrapulmonary SFTs who underwent surgical resection at the Department of Cardiothoracic Surgery, Lishui Center Hospital (Lishui, China), and Clinical College of Yangzhou University, (Yangzhou, China), between January 2000 and

January 2016. Age, sex, medical history, clinical presentation, diagnostic methods, intraoperative findings, postoperative complications and outcome were retrieved from hospital records. Meanwhile, relevant studies regarding intrapulmonary SFTs were searched via PubMed from January 1990 to January 2016. The text words and MeSH terms 'Solitary fibrous tumours', 'Intrapulmonary', and 'Lung' were used. Tumour characteristics, clinicopathologic features, therapeutic strategy and survival outcomes were reviewed, and these data were tabulated.

Results

Report of cases. Of the five cases, all were males, with a mean age of 57.6 years (range, 37-68 years). Two patients (nos. 2 and 5) had history of hypertensive disease. One patient (no. 2) had history of diabetes mellitus. One patient (no. 1) had history of bronchial asthma. One patient (no. 3) had history of nodular goitre. The remaining patient (no. 4) had no history of any disease. All patients were asymptomatic, and their tumours were discovered incidentally on routine computed tomography (CT) examination. Contrast-enhanced CT of the chest revealed a lung mass with no calcification or any fatty tissue (Fig. 1). One, one, one and two tumours occurred in the right lower, left upper, right upper and left lower lung, respectively. Three patients (nos. 1-3) were preoperatively diagnosed with spindle cell tumour by CT-guided percutaneous aspiration biopsy. Other examinations, including pulmonary function, echocardiogram, electrocardiogram, coagulation function and blood routine examination, were normal. The serum levels of Na^+, K^+, Cl^-, Ca^{2+}, Mg^{2+} were all within reference range. Serum carbohydrate antigen, carcino-embryonic antigen, squamous cell antigen and neuron-specific enolase were within normal limits. No evidence of metastasis was found via head magnetic resonance imaging (MRI) and abdominal ultrasound.

One patient underwent tumour enucleation though thoracotomy (no. 1), one patient underwent upper left lobectomy associated with lymph node dissection involving radical dissection of the mediastinum (no. 2), and one patient underwent tumour resection associated with bilateral subtotal thyroidectomy (no. 3). The remaining two patients underwent tumour enucleation via video-assisted thoracoscopic surgery (VATS) (nos. 4 and 5) (Figs. 2 and 3). Data of the clinical features are shown in Table I.

The haematoxylin and eosin (H&E) stain showed a rich variety of spindle cells and amorphous areas of collagen (Fig. 4). Immunohistochemical reactions of the tumour cells were strongly positive for B-cell lymphoma (Bcl)-2 (Fig. 5) and cluster of differentiation (CD)34 (Fig. 6). Tumour cells were positive for vimentin (nos. 4 and 5) (Fig. 7) and CD99 (nos. 2-5) (Fig. 8). The tumour cells were negative for CD117, desmin, smooth muscle actin (SMA), epithelial membrane antigen and S-100. Following an immunohistochemical-analysis-based study, the diagnosis of intrapulmonary SFTs was made.

Postoperative haemorrhage occurred in one patient (no. 1), and he received surgical intervention for haemostasis. The average time of intrathoracic drain was 3.6 (1-9) days. The average hospital stay was 15 (4-22) days, and

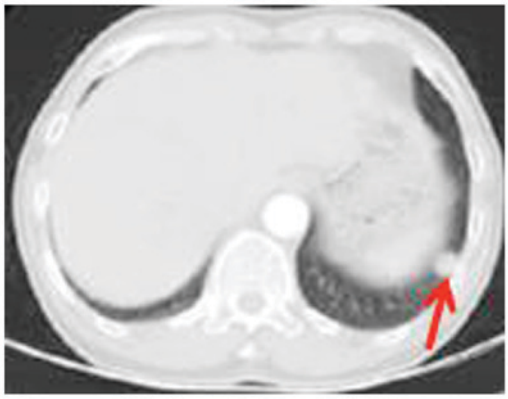

Figure 1. CT scan of the chest demonstrated a 1.5x1.5 cm left lung mass (red arrow). CT, computed tomography.

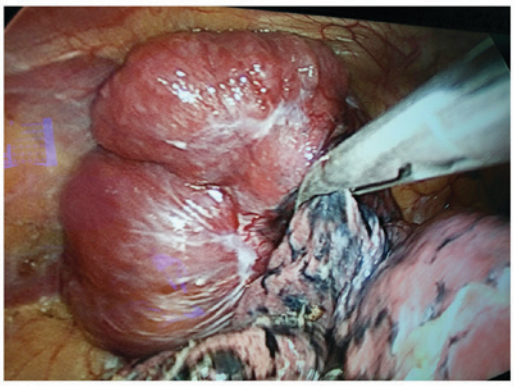

Figure 2. Surgical procedure, partially dissected right upper lung mass.

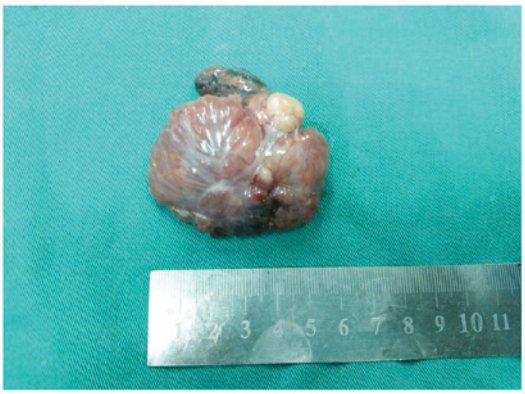

Figure 3. Surgical specimen of 5.5x5x5 cm and weighing 325 g.

no mortality occurred. All patients were discharged from the hospital following an uneventfully recovery. The mean length of the postoperative follow-up was 37.6 (1-67) months. One patient was lost to follow-up, and four patients were asymptomatic.

Published case study findings. Nineteen articles were identified from the searches of databases (17,43-59) (Table II). They had a total of 45 patients: Twenty-three males and 22 females. The mean age was 59.4 years, ranging from 7 to 81 years. Twelve patients were asymptomatic, and pain

Table I. Patient characteristics and treatment history in our study.

Features	Patient no. 1	Patient no. 2	Patient no. 3	Patient no.4	Patient no.5
Sex	Male	Male	Male	Male	Male
Age (years)	37	59	59	65	68
Presentation	Asymptomatic	Asymptomatic	Asymptomatic	Asymptomatic	Asymptomatic
Previous history	Bronchial asthma	Hypertensive disease, diabetes mellitus	Nodular goiter	No	Hypertensive disease
Paraneoplastic syndrome	No	No	No	No	No
Location of lesions	Right lower lobe	Left lower lobe	Left upper lobe	Left lower lobe	Right upper lobe
Surgical strategy	Thoracotomy	Thoracotomy	Thoracotomy + bilateral subtotal thyroidectomy	VATS	VATS
Surgical procedures	Adequate wedge resection	Left lower lobectomy associated with lymph node dissection	Left upper lobectomy	Adequate wedge resection	Adequate wedge resection
Operating time (min)	100	120	150	75	35
Blood loss (ml)	200	300	100	5	50
Size of lesion (cm)	2.5x3.0	7x5	6.5x3	1.5x1.5	4x3
Postoperative complication	Postoperative hemorrhage	no	No	No	no
Hospital stay (days)	22	18	22	4	9
Time of intrathoracic drain (days)	9	5	1	1	2
Cellular pattern	Spindle	Spindle	Spindle	Spindle	Spindle
Mitotic count	<1/10HPF	>10/10HPF	<5/10HPF	<1/10HPF	<5/10HPF
CD34	+	+	+	+	+
CD99	-	+	+	+	+
Bcl-2	+	+	+	+	+
Vimentin	-	-	-	+	+
Desmin	-	-	-	-	-
S-100	-	-	-	-	-
SMA	-	-	-	-	-
Diagnosis	Benign	Malignant	Benign	Benign	Benign
Follow up (months)	38	67	55	27	1
Recurrence	Unknown	No	No	No	No
Present status	Unknown	NED	NED	NED	NED

NED, no evidence of disease; VATS, video-assisted thoracoscopic surgery; CD, cluster of differentiation; Blc-2, B-cell lymphoma 2; SMA, smooth muscle actin.

and cough were the major symptoms. Five, one, two, four, and seventeen tumours occurred in the right upper lobe, right middle lobe, right lower lobe, left upper lobe and left lower lobe, respectively. Thirty-nine patients underwent surgery, one patient underwent radiotherapy, and one patient underwent radiofrequency ablation. Twenty-two patients were followed up, and the mean length of the post-operative follow-up was 48 (1-168) months. One patient was diagnosed with chest wall metastases. Five patients died (Table III).

Pathologic and immunohistochemical features. Twenty-eight of 32 cases showed low mitotic counts (0-5/10 HPF), 1 of 32 had a middle mitotic count (0-6/10 HPF), and 3 of 32 showed high mitotic counts (5-10/10 HPF). Immunohistochemical staining analyses were performed in 28 cases. Twenty-four of 28 cases were CD34-positive. Ten of 13 cases were CD99-positive. Fourteen of 15 cases were Bcl-2 positive. Fourteen of 16 cases were vimentin positive. Five of 17 cases were smooth muscle antibody (SMA)-positive. Two of 9 cases were epithelial membrane antigen (EMA)

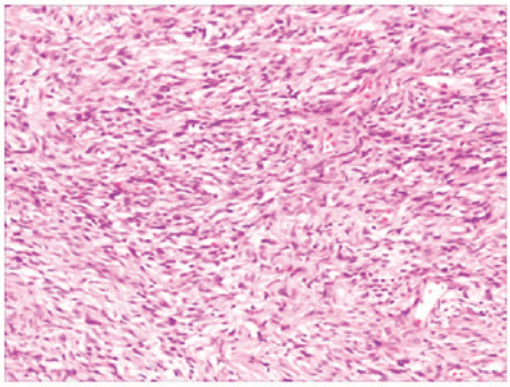

Figure 4. Haematoxylin and eosin staining showed a rich variety of spindle cells and amorphous areas of collagen (magnification, x100).

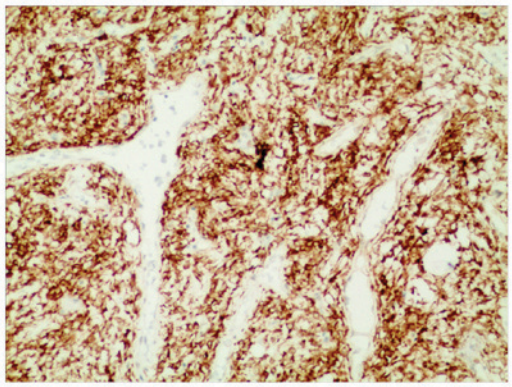

Figure 5. The immunohistochemical reactions for Bcl-2 were positive (magnification, x100). Bcl-2, B-cell lymphoma-2.

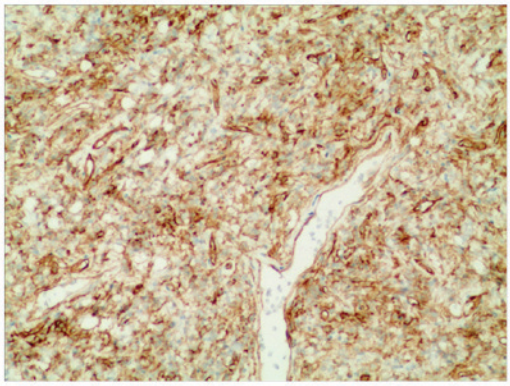

Figure 6. The immunohistochemical reactions for CD34 protein were positive (magnification, x100). CD, cluster of differentiation.

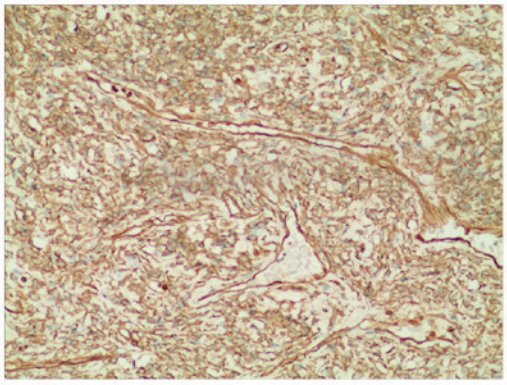

Figure 7. The immunohistochemical reactions for vimentin protein were positive (magnification, x100).

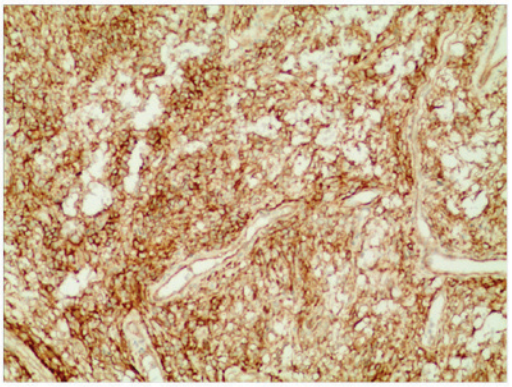

Figure 8. The immunohistochemical reactions for CD99 protein were positive (magnification, x100). CD, cluster of differentiation.

positive. Eight of 9 cases were mib-monoclonal antibody-1 (MIB-1)-positive (Table IV).

Discussion

SFTs are rare, mesenchymal neoplasms initially described in the pleura but have since been discovered in nearly every anatomic location (61). Klemperer and Rabin (62) reported 5 cases of primary pleural neoplasms in 1931 and proposed that SFT was of submesothelial origin. However, in the subsequent decades, on the basis of immunohistochemical analyses and ultrastructural features, it is now recognized that SFTs arise from primitive fibroblast-like cells in connective tissue (61). SFTs, to our knowledge, most often occur in the pleura. They can be rarely found in the lung, central nervous system, kidney and other extrapleural sites. Via searches of databases, a total of 45 patients with intrapulmonary SFTs were found.

Intrapulmonary SFTs are usually found incidentally and may be associated with chest pain and cough. Seventeen patients (5 in our study and 12 in published literature) were asymptomatic and were discovered incidentally. Two patients presented with pain, and two patients presented with cough. Of note, few patients with SFTs present with refractory hypoglycaemia, which is a paraneoplastic syndrome that secretes a prohormone form of insulin-like growth factor-II (IGF-II), referred to as the Doege-Potter syndrome (DPS).

Due to their atypical clinical and radiographic appearance as a common lung tumour, the diagnosis of intrapulmonary SFTs presents unique challenges. Imaging examinations, including chest X-rays, CT and MRI, are used for assessing intrapulmonary SFTs. There are limited data on X-rays and CT imaging features of intrapulmonary SFTs. The available data show that intrapulmonary SFTs are well-defined ovoid or round pulmonary nodules on chest X-rays and CT scanning, but they are non-specific. The PET-CT findings of SFTs have

Table II. Clinicopathologic features of intrapulmonary solitary fibrous tumours present in the English literature.

Authors (ref.)	Case	Age (years)	Sex	Site	Size	Symptoms	Treatment	Follow-up (months)	Status
Cardinal e*t al* (43)	3	44	Male	Left lower lobe	4 cm	Chest discomfort	Segmental resection	48	NED
		64	Male	Left lower lobe	6 cm	Asymptomatic	Left lower lobectomy	36	NED
		47	Male	Left lower lobe	3 cm	Asymptomatic	Tumor resection	12	NED
Ikeda *et al* (44)	1	80	Female	Left lower lobe	Unknown	Back pain	Radiotherapy	11	Died
Sironi *et al* (45)	1	68	Male	Left lower lobe	8.8x5.7x5 cm	Asymptomatic	Unknown	Unknown	Unknown
Dong *et al* (46)	1	18	Male	Bilateral lungs	Unknown	Asymptomatic	Unknown	Unknown	Unknown
van de Rijn *et al* (47)	2	69	Male	Unknown	12 cm	Unknown	Autopsy	No	Died
		80	Female	Unknown	Unknown	Unknown	Unknown	79	NED
Demırağ *et al* (48)	1	56	Female	Left lung	16 cm	Unknown	Left pneumonectomy	87	Unknown
Barrettara *et al* (49)	1	81	Female	Left lower lobe	10x9 cm	Left thoracic pain	Left inferior lobectomy	Unknown	Unknown
Patsios *et al* (50)	1	50	Male	Left lower lobe	2.7 cm	Asymptomatic	Wedge resection	Unknown	Unknown
Sakurai *et al* (51)	1	40	Male	Left lower lobe	2.2 cm	Asymptomatic	Wedge resection	14	NED
Sagawa *et al* (52)	1	72	Female	Left upper lobe	12x9x7 mm	Asymptomatic	Wedge resection	12	NED
Geramizadeh *et al* (53)	1	7	Male	Right upper lobe	5 cm	Cough and dyspnea	Right pneumonectomy	9	NED
Kawaguchi *et al* (17)	1	60	Female	Left upper lobe	23x22x19 mm	Asymptomatic	Left upper segmentectomy	6	NED
Rao *et al* (54)	24	83	Male	Right lung	13.0 cm	Unknown	Wedge excision or lobectomy	<60	NED
		75	Female	Unknown	Unknown			<60	NED
		73	Female	Left lower lobe	2.3 cm			<60	NED
		69	Male	Left lower lobe	Unknown			<60	NED
		59	Female	Unknown	Unknown			<60	NED
		52	Female	Left upper lobe	2.5 cm			108	NED
		49	Male	Unknown	18.0 cm			168	NED
		58	Female	Left lower lobe	3.9 cm			<60	NED
		46	Male	Right lung	4.5 cm			156	NED
		64	Male	Right upper lobe	5.0 cm			84	Died
		68	Female	Left lung	Unknown			Unknown	Unknown
		60	Male	Left lower lobe	Unknown			Unknown	Unknown
		59	Male	Right upper lobe	Unknown			Unknown	Unknown
		44	Female	Right upper lobe	Unknown			Unknown	Unknown
		50	Female	Left lower lobe	Unknown			Unknown	Unknown
		81	Female	Unknown	7.0 cm			Unknown	Unknown
		61	Male	Right lower lobe	22.0 cm			48	Died
		64	Female	Left lower lobe	12.0 cm			<60	NED
		62	Male	Unknown	8.0 cm			<60	NED

Table II. Continued.

Authors (ref.)	Case	Age (years)	Sex	Site	Size	Symptoms	Treatment	Follow-up (months)	Status
		44	Female	Left lung	4.5 cm			<60	NED
		73	Female	Unknown	3.5 cm			<60	NED
		75	Female	Unknown	10.0 cm			60	Chest wall metastases
		59	Female	Left lung	10.0 cm			Unknown	Unknown
		45	Male	Left lower lobe	3.0 cm			60	Died
Caruso *et al* (55)	1	72	Male	Left lower lobe	6.0 cm	Asymptomatic	Left lower lobectomy	12	NED
Fridlington *et al* (56)	1	56	Male	Left lower lobe	20 cm	Symptomatic hypoglycemia	Pneumonectomy	1	NED
Kouki *et al* (57)	1	52	Male	Right upper lobe	5.3x5.0 cm	Asymptomatic	Right upper lobectomy	24	NED
Baliga *et al* (58)	1	42	Male	Right lower lobe	11 cm	Asymptomatic	Radiofrequency ablation	No	Died
Khalifa *et al* (59)	1	71	Female	Right middle lobe	2.8 cm	Presistent cough with clear sputum	Wedge lung resection	10	NED
Chang *et al* (60)	1	73	Female	Left lower lobe	3x3x2 cm	Asymptomatic	Lobectomy	12	NED

NED, no evidence of disease.

Table III. Characteristics of the primary intrapulmonary solitary fibrous tumors present in the English literature.

No. of studies	19
No. cases	45
Age (years)	
Mean	59.4
Range	7-81
Tumor sizes (cm)	
Mean	8.2
Range	2-23
Sex	
Male	23 (51.1%)
Female	22 (48.9%)
Symptoms	18
Asymptomatic	12 (66.7%)
Pain	2 (11.1%)
Cough	2 (11.1%)
Other	2 (11.1%)
Localization	29
Right upper lobe	5 (17.2%)
Right middle lobe	1 (3.5%)
Right lower lobe	2 (6.9%)
Left upper lobe	4 (13.8%)
Left lower lobe	17 (58.6%)
Treatment	41
Surgery	39 (95.2%)
Radiotherapy	1 (2.4%)
Radiofrequency ablation	1 (2.4%)

been rarely reported. PET-CT is a useful tool for evaluating the size, regional invasion and distant metastasis of a tumour. Yan *et al* (63) have reported a malignant SFT with mildly increased FDG uptake. Dong *et al* (46) have reported a benign SFT with intense FDG uptake. The clinical behaviour of the tumour may be predicted based on PET-CT findings, and FDG uptake degree may be related to the tumour's aggressive behaviour (46). An intrapulmonary SFT may be identified by CT-guided percutaneous aspiration biopsy. Caruso *et al* (55) operated on a pulmonary mass CT-guided FNA cytology biopsy. The FNA cytologic specimen contained spindle cells. Furthermore, FNA cytology, proper clinical and radiologic findings are helpful in narrowing the diagnostic possibilities and making tentative pathologic diagnoses. However, spindle-shaped cells also appear in fibrosarcoma, leiomyosarcoma, schwannoma and others. Therefore, the diagnosis of SFTs may not be identified without immunohistochemical staining. The differential diagnosis of intrapulmonary SFT includes numerous malignant and benign tumours, including pulmonary adenofibroma, benign neural neoplasms, leiomyoma, leiomyosarcoma, synovial sarcoma, spindle cell thymoma, spindle cell carcinoid tumour, nerve sheath tumour, fibrosarcoma, sarcomatoid carcinoma, and sarcomatoid mesothelioma (54). To our knowledge, positive CD34,

Table IV. Pathologic and immunohistochemical features presented in the English literature.

Author	Mitotic count	Immunohistochemistry
Sironi *et al* (45)	NA	CD34^{+}, CD99^{+}
Dong *et al* (46)	NA	Vimentin^{+}, CD34^{+}, EMA^{-}, CAM5.2^{-}, S-100^{-}, SMA^{-}, desmin^{-}, HMB45^{-}
van de Rijn *et al* (47)	NA	CD34^{+}, SMA^{-}, MSA^{-}, desmin^{-}, AE1^{-}, CAM5^{-}
Demırağ *et al* (48)	2/10 HPF	CD44^{+++}, MMP-2^{+}
Barrettara *et al* (49)	2/10 HPF	CD34^{+}, CD99^{+}, Bcl-2^{+}, vimentin^{+}, calretinin^{-}, S100^{-}, actine^{-}, CK^{-}
Patsios *et al* (50)	NA	Vimentin^{+}, CD34: focal^{+}, CK^{-}, S-100^{-}, SMA^{-}
Sakurai *et al* (51)	NA	Vimentin^{+}, CD34^{+}, Bcl-2^{+}, CK^{-}, desmin^{-}, SMA^{-}, S-100^{-}
Sagawa *et al* (52)	Rare	CD34^{+}, TTF-1^{-}, MIB-1^{-}
Geramizadeh *et al* (53)	0/10 HPF	Vimentin^{+}, CD34^{+}, Bcl-2^{+}, CK^{-}, desmin^{-}, SMA^{-}, S-100^{-}
Kawaguchi *et al* (17)	NA	CD34^{+}, calretinin^{-}, SMA^{-}, S-100^{-}
Rao *et al* (54)	2-5/10 HPF	NA
	2-5/10 HPF	Vimentin^{+}, p53^{+}, Bcl-2^{+}, CD34^{+}, CD99^{+}, MIB-1^{++}, CK AE1/AE3^{-}, EMA^{-}, S100^{-}, SMMS-1^{-}
	<1/10 HPF	NA
	<1/10 HPF	NA
	<1/10 HPF	NA
	<1/10 HPF	NA
	2-5/10 HPF	Bcl-2^{+}, CD34^{+}, CD99^{+}, MIB-1^{+}, SMA^{+}, vimentin^{-}, p53^{-}, calponin^{-}, AE1/3^{-}, EMA^{-}, S100^{-}, SMMS1^{-}
	<1/10 HPF	Bcl-2^{+}, CD34^{+}, CK AE1/3^{-}, SMA^{-}
	2-5/10 HPF	NA
	<1/10 HPF	CD34^{+}, CD99^{+}, MIB-1^{+}, SMA^{-}, CK AE1/3^{-}
	<1/10 HPF	NA
	2-5/10 HPF	NA
	<1/10 HPF	NA
	<1/10 HPF	NA
	<1/10 HPF	NA
	<1/10 HPF	NA
	2-5/10 HPF	Vimentin^{+++}, Bcl-2^{+++}, calponin^{++}, CD34^{++}, CD99-weak^{+}, CK AE1/3: focal^{+}, EMA^{-}, MIB-1^{+}, SMA^{-}
	<1/10 HPF	Bcl-2^{+++}, CD34^{+}, CD99^{+}, MIB-1^{+}, SMA^{+}, CK AE1/3^{-}, vimentin^{-}, calponin^{-}, EMA^{-},
	2-5/10 HPF	Vimentin^{+++}, Bcl-2^{+++}, CD34^{+++}, MIB-1^{++}, CD99^{++}, calponin^{+}, CK AE1/3^{-}, EMA^{-}, S100^{-}, SMA^{-}, SMMS1^{-}
	<1/10 HPF	CD99^{+++}, Bcl-2^{+++}, CD34^{+}, p53^{+}, MIB-1^{+}, calponin^{-}, AE1/3^{-}, EMA^{-}, S100^{-}, SMMS1^{-}
	<1/10 HPF	CD99^{-}, Bcl-2^{+++}, CD34^{++}, p53^{+}, AE1/3^{-}, SMA^{-}, vimentin^{+++}, SMMS1^{-}
	5-10/10 HPF	Bcl-2^{+}, CD34^{+}, CD99^{+}, SMA^{+}, CK AE1/3^{-}
	>10/10 HPF	Vimentin^{+}, Bcl-2^{+}, CD99^{+}, p53^{-}, calponin^{-}, CD34^{-}, CK AE1/3^{-}, S100^{-}, SMA^{-}, SMMS1^{-}
	>10/10 HPF	MIB-1^{+++}, SMA^{++}, EMA^{+}, p53^{-}, Bcl-2^{-}, calponin^{-}, CD34^{-}, CD99^{-}, CK AE1/3^{-}, S100^{-}, SMMS1^{-}
Caruso *et al* (55)	0-6/10 HPF	Vimentin^{+}, keratin^{-}, CEA^{-}, EMA^{+}, F VIII^{-}, S-100^{-}, desmin^{-}, actin^{-}
Fridlington *et al* (56)	1-2/10 HPF	CD34^{+}, vimentin^{+}, S-100^{-}
Kouki *et al* (57)	3-4/10 HPF	CD34^{+}
Baliga *et al* (58)	0/10 HPF	CD34^{++}, Bcl-2^{+}, SMA^{+}, CD99^{-}, CAM 5.2^{-}, calretinin^{-}
Khalifa *et al* (59)	NA	CD34^{+}, vimentin^{+}, S-100-, CK^{-}, F VIII^{-}, MSA^{-}, SMA^{-}
Chang *et al* (60)	NA	CD34^{+}, vimentin^{+}, S-100^{-}, desmin^{-}, CEA^{-}, a1-ACT^{-}, F VIII^{-}

HPF, high power field; CD, cluster of differentiation; Bcl, B-cell lymphoma; EMA, epithelial membrane antigen; CAM, low molecular weight cytokeratin; CEA, carcinoembryonic antigen; MMP, matrix metalloproteinase; SMMS, smooth muscle myosin; CK, creatine kinase.

CD99, vimentin and Bcl-2 expression are important markers for the diagnosis of an SFT.

Our review of previously reported cases in the literature suggests complete surgical resection is generally accepted

as the definitive and effective treatment of choice for intrapulmonary SFTs. Adequate wedge resection, anatomic segmentectomy and lobectomy, according to the location of mass, are common surgical procedures for resection of intrapulmonary tumours. In our study, three patients underwent adequate wedge resection, whereas the other two patients under anatomic lobectomy. In recent decades, with the development of minimally invasive technology, surgical approaches have been radically changed. More and more intrapulmonary tumours can be safely and availably excised via VATS. In our study, two patients underwent VATS. Previous studies have used radiotherapy and radiofrequency ablation for intrapulmonary SFTs. However, these treatments carry a higher risk of death during follow-up. The effectiveness and capability of such treatment has yet to be confirmed.

Due to their deficiency of information and their languages of publication (not English), some studies were not included in our study. Thirty-six of 681 cases and 8 of 88 cases were SFTs of the lung and pleura, respectively, in Choi's *et al* (64) and Schirosi's *et al* (65) studies, and these were not included. Gonullu *et al* (66) reported a case of metastatic breast carcinoma to SFT in the lung, and Strickland *et al* (67) reported a case of SFT of the uterus presenting with lung metastases. Míguez González *et al* (68) reported a case of intrapulmonary SFT associated with haemoptysis, and Masuda *et al* (69) report two cases of intrapulmonary tumours with different growth patterns. However, the articles were not written in English. Radulescu *et al* (70) reported a case of malignant primary intrapulmonary SFT with haemangiopericytoma-like features.

In conclusion, we retrospectively reviewed the records of 5 patients with primary intrapulmonary SFTs who underwent surgical resection, and we systematically reviewed previously reported cases in the literature. This study has 4 important findings. First, intrapulmonary SFTs can occur in people of all ages and with no sex predominance. The male-to-female ratio was 1.0:0.96. Second, intrapulmonary SFTs are often discovered incidentally. When symptomatic, the common symptoms are chest pain and cough. Third, the left lower lobe was the most common site location. Finally, complete surgical resection is a safe, effective and successful treatment.

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