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Distant Metastasis from Benign Solitary Fibrous Tumor of the Kidney

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Key Words
Solitary fibrous tumor · Metastasis · Kidney · Malignant · CD34

Abstract
Solitary fibrous tumor (SFT) rarely occurs in the kidneys, and only one reported case of renal SFT has shown distant metastasis. We report the second case of renal SFT exhibiting distant metastasis. A 48-year-old man was referred to our hospital because of a right renal mass. An abdominal CT scan detected a large renal tumor, which was suspected to be a renal cell carcinoma. Right radical nephrectomy was performed, and the tumor was found to measure 28 × 18 × 10 cm. The pathological diagnosis was benign solitary fibrous tumor of the kidney. Eight years after the operation, lung and liver metastases developed. Pulmonary segmentectomy and partial hepatectomy were performed. The pathological diagnoses of these resected tissue specimens were compatible with benign SFT.

Introduction

Solitary fibrous tumor (SFT) was first reported by Klemperer and Rabin in 1931 as a tumor of the pleura [1]. SFT is a rare type of spindle cell neoplasm that usually arises in the pleura [2]. However, it has also been reported to occur at other sites. SFT of the kidneys is rare, and furthermore, distant metastasis from SFT is extremely rare. We report the second case of renal SFT to exhibit distant metastasis.

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Case Report

A 48-year-old man was referred to our hospital because of a right renal mass. A physical examination revealed a hard right abdominal mass, and a subsequent CT scan detected a large right renal tumor, which was suspected to be a renal cell carcinoma (fig. 1). The patient underwent radical right nephrectomy. In gross appearance, the tumor measured 28 × 18 × 10 cm, and displayed cystic changes, necrosis and hemorrhage with grayish-white cut surfaces. The tumor developed from the upper pole of the kidney adjacent to the renal capsule and markedly compressed the normal kidney into the lower side.

Microscopically, the tumor was found adjacent to the renal capsule; but the site of origin was ambiguous. It consisted of spindle-shaped cells with scant cytoplasm accompanied by prominent hyalinized collagenous tissue, which displayed hemangiopericytomatous patterns. The cells did not display cytological atypia, and no mitotic figures were detected. Immunohistochemical staining was positive for CD34, vimentin, and CD99 and negative for keratin, EMA, CD10, CD31, factor VIII, αSMA, Bcl-2, S-100, and CD117 (fig. 2). These findings resulted in a diagnosis of benign SFT.

However, 8 years after the original operation, follow-up CT detected a lung nodule and multiple liver nodules (fig. 3), which were consistent with metastasis from the primary renal SFT. Ultrasound-guided liver needle biopsy was performed and led to a pathological diagnosis of SFT. Surgical resection was planned to treat the tumors, and partial hepatectomy was performed. Two months after the partial hepatectomy, pulmonary segmentectomy was performed. Microscopically, the metastatic tumors were composed of spindle cells in a collagenous stroma containing hemangiopericytomatous structures. The cells did not display cytological atypia, and no mitotic figures were detected. Although immunohistochemical staining for CD34 was negative in the resected tissue from the liver and weakly positive in the resected tissue from the lungs, the specimens were positively stained for vimentin and CD99 and negative for CD10, factor VIII, αSMA, Bcl-2, and S-100 (fig. 2). The pathological diagnosis was benign SFT.

As the CD34 expression of the primary tumor and metastatic lesion differed, we re-evaluated the pathological findings of the primary renal SFT. Immunohistochemically, we observed CD34 labeling in 55% of the tumor cells with no expression in the remaining component. The CD34-negative section of the lesion was morphologically indistinguishable from the CD34-positive part of the lesion. Pathologically malignant findings were not observed in any of the lesions. At 12 months after surgery, the patient is healthy and has not displayed any evidence of recurrence or metastasis.

Discussion

SFT was first reported by Klemperer and Rabin in 1931 as a tumor of the pleura [1]. SFT is a rare spindle cell neoplasm that usually arises in the pleura [2]. However, in recent years, there have been several reports of SFT arising in other organs, including the kidneys [2, 3]. The histogenesis of SFT has been debated for years, but recent studies have indicated that it has a mesenchymal origin [4]. Immunohistochemical studies are useful for establishing a diagnosis, especially staining for CD34, which is considered to be a marker of SFT [5]. In addition, most SFT are diffusely positive for Bcl-2 and CD99 [6]. Loss of Bcl-2 was closely related to high malignant potential in extrathoracic SFT [7]. Surgical resection has been demonstrated to be beneficial in the treatment of SFT. Even if the SFT is histologically

diagnosed as malignant, complete excision of the tumor is associated with a favorable prognosis [8].

SFT of the kidneys is rare, and only 67 cases have been reported (table 1). Most of these tumors were preoperatively diagnosed as renal cell carcinoma, and radical nephrectomy was the standard treatment. Pathologically, 61 tumors were diagnosed as benign and 6 tumors were diagnosed as malignant. All tumors except one displayed a favorable prognosis, with no evidence of recurrence during the follow-up period, which ranged from 2 to 89 months. Immunohistochemically, most tumors were positive for CD34. Although the origin of renal SFT is difficult to determine, some reported renal SFT originated from the renal capsule [3], and Yamada et al. [9] speculated that renal SFT originates from primitive mesenchymal cells located in the renal capsule. Further research is necessary to clarify the origin of renal SFT.

Fine et al. [10] reported the first case of malignant renal SFT to develop distant metastasis. Their case involved a 76-year-old man who was treated with left radical nephrectomy. Pathologically, 10% of the renal tumor consisted of typical benign SFT; however, the remaining component was composed of pleomorphic, spindle-shaped sarcoma cells with frequent mitoses and necrotic foci. Immunohistochemically, CD34 labeling was observed in the benign SFT component with no CD34 expression in the sarcomatous component. Four months after surgery, multiple lung metastases developed. This was the first reported case of malignant renal SFT involving distant metastasis. In this patient, neither metastasectomy nor a histological examination of the metastatic lesion was performed.

To the best of our knowledge, our case is the second reported case of renal SFT to involve distant metastasis and is the first reported case of renal SFT to include the pathological findings of the metastatic lesion. Furthermore, the primary tumor and the resected tissue from the metastatic site were pathologically benign, and no malignant findings were observed in any of the lesions. Immunohistochemically, 55% of the primary tumor displayed positive CD34 labeling, whereas no CD34 expression was detected in the remaining component. The CD34-negative part of the lesion was morphologically indistinguishable from its CD34-positive region. In addition, CD34 expression was negative in the resected tissue from the liver and weakly positive in the resected tissue from the lungs. Thus, we postulated that the loss of CD34 expression might promote tumor metastasis to other organs, and could lead to malignant transformation from the benign tumor relevant to fatal outcome [11]. Moreover, our case of SFT was negative in Bcl-2 expression in the primary tumor and metastatic lesions; this may also participate in malignant outcome [7]. Further research is needed to clarify these points.

Our case is very similar to that reported by Hasegawa et al. [12]. They described an extrathoracic SFT that metastasized to the lungs. Neither the primary nor metastatic lesions displayed any atypical features. Thus, extrathoracic SFT might have the potential to recur or metastasize, even in the absence of atypical pathological features [12]. Renal SFT is generally reported to be a benign tumor; however, the follow-up periods in the 67 reported cases might not have been sufficient to allow the clinical outcome to be fully evaluated (table 1). A longer follow-up period might be necessary to definitively evaluate the clinical outcome of renal SFT.

Disclosure Statement

The authors have no conflicts of interest to disclose.

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Table 1. Clinicopathological findings of renal solitary fibrous tumors in the literature

Case	Year	Age years	Sex	Symptom	Side	Affected site	Tumor size, cm	Treatment	Histology	Follow-up	Outcome	CD34*	Authors and journals
1	1996	48	M	Back pain and macrohematuria	R	Renal capsule	3	Nephrectomy	BEN	0.1	DNOD	POS	Gelb et al., Am J Surg Pathol 20:1288
2	1996	45	F	Incidental	R	Kidney	6	Nephrectomy	BEN	8	NED	POS (2/3)	Fain et al., J Urol Pathol 4:227
3	1996	46	F	Incidental	R	Kidney	7.2	Nephrectomy	BEN	33	NED	POS (2/3)	Fain et al., J Urol Pathol 4:227
4	1996	51	M	Incidental	L	Kidney	4.5	Nephrectomy	BEN	2	NED	POS (2/3)	Fain et al., J Urol Pathol 4:227
5	1997	33	F	Abdominal pain	R	Peripelvis	3.5	Nephrectomy	BEN	89	NED	POS	Fukunaga et al., Hispathology 30:451
6	1997	36	F	Abdominal pain	L	Peripelvis	2	Nephrectomy	BEN	12	NED	POS	Fukunaga et al., Hispathology 30:451
7	1998	59	M	Incidental	L	Renal capsule	NA	Nephrectomy	BEN	NA	NA	POS	Ookouci S et al., Jpn J Radiol 58: 539
8	1998	57	M	Incidental	L	Kidney	7	Tumorectomy	BEN	NA	NA	POS	Tanahashi C et al., Proc Jpn Soc Pathol 87:510
9	1999	64	M	Macrohematuria	R	Kidney	4.5	Nephrectomy	BEN	8	NED	POS	Hasegawa et al., Hum Pathol 30:1464
10	1999	71	F	Incidental	L	Kidney	9	Nephrectomy	BEN	NA	NA	NA	Kojima K et al., Jap-Deu Med Beriche 44:185
11	2000	66	F	Abdominal pain and macrohematuria	R	Kidney	9	Nephrectomy	BEN	9	NED	POS	Leroy et al., Urol Int 65:49
12	2000	72	F	NA	L	Kidney	8	Nephrectomy	BEN	10	NED	POS	Morimitsu et al., APMIS 108:617
13	2000	56	F	Incidental	L	Renal capsule	5	Tumor resection	BEN	NA	NA	NA	Ikeda A et al., J Hiroshima Med Assoc 53:640
14	2001	70	M	Incidental	R	Renal pelvis	6	Nephrectomy	BEN	60	NED	POS	Yazaki et al., Int J Urol 8:504
15	2001	28	F	Abdominal pain	L	Kidney	15	Nephrectomy	BEN	12	NED	POS	Cortes-Gutierrez et al., J Urol 166:60
16	2001	41	M	Macrohematuria	L	Kidney	14	Nephrectomy	BEN	48	NED	POS	Wang J et al., Am J Surg Pathol 25:1194
17	2001	72	M	Abdominal discomfort	R	Kidney	13	Nephrectomy	BEN	5	NED	POS	Wang J et al., Am J Surg Pathol 25:1194
18	2002	57	M	Incidental	L	Kidney	6	Nephrectomy	BEN	NA	NA	POS	Miyazaki N et al., Jpn Red Cross Med J 54:182
19	2002	58	M	Incidental	L	Kidney	NA	Nephrectomy	BEN	9	NED	NA	Inokawa E, J Hiroshima Med Assoc 55:1057
20	2002	31	F	Flank pain	R	Kidney	8.6	Nephrectomy	BEN	8	NED	POS	Magro G, Pathol Res Pract 198:37
21	2003	64	F	Microhematuria	R	Kidney	4	Nephrectomy	BEN	7	NED	POS	Li S et al., Hinyokika Kiyo 49:121
22	2003	51	F	NA	R/L	Kidney	25 & 2	Tumor resection	BEN	NA	NA	NA	Llarena Ibarguren et al., Arch Esp Urol 56:835
23	2003	35	M	NA	R	Kidney	17	Nephrectomy	BEN	6	NED	NA	Durand X et al., Prog Urol 13:491
24	2003	60	F	NA	R	Kidney	11	Nephrectomy	BEN	48	NED	NA	Bugel H et al., Prog Urol 13:1397
25	2004	67	M	Incidental	L	Kidney	4.5	Tumorectomy	BEN	5	NED	POS	Toriyama S et al., Hinyokika Kiyo 50:138
26	2004	83	M	NA	R	Kidney	9	Nephrectomy	BEN	18	NED	POS	Gres P et al., Prog Urol 14:65
27	2004	53	M	Flank pain and swelling	R	Renal capsule	14	Tumor resection	BEN	36	DNOD	POS	Kunieda K et al., Surg Today 34:90
28	2004	59	M	Incidental	L	Renal capsule	6.8	Nephrectomy	BEN	48	NED	POS	Yamada H et al., Pathol Int 54:914
29–35	2005	29–79	NA	5 incidental and 2 flank pain	NA	6 renal, 1 perirenal	2.2–10.1	Nephrectomy	BEN	NA	NA	POS in 6	Pierson DM et al., Mod Pathol 18:159
36	2005	51	F	Flank pain	NA	Renal capsule	10	Nephrectomy	BEN	NA	NA	POS	Yamaguchi T, Urology 65:175
37	2005	51	F	Fever elevation	R	Renal capsule	13	Nephrectomy	BEN	NA	NA	POS (focal)	Jhonson TR et al., J Comput Assist Tomogr 29:481
38	2005	83	F	Incidental	L	Kidney	11	Nephrectomy	BEN	NA	NA	POS	Kawagoe M, Nishinihon J Urol 67:568
39	2006	76	M	Incidental	L	Kidney	12	Nephrectomy	MAL	4	Lung metastasis	POS (benign site)	Fine SW et al., Arch Pathol Lab Med 130:857
40	2006	18	F	Flank pain	L	Kidney	3	Nephrectomy	BEN	15	NED	POS	Koroku M et al., Hinyokika Kiyo 52:705
41	2006	4	M	NA	R	Kidney	8	Nephrectomy	BEN	NA	NA	NA	Provance et al., Clin Pediatr 45:871
42	2006	85	M	Flank pain	L	Kidney	4.5	Nephrectomy	BEN	NA	NA	POS	Kohl SK et al., Arch Pathol Lab Med 130:117
43	2006	54	M	Incidental	R	Kidney	NA	Nephrectomy	BEN	16	NED	POS	Tanaka M et al., Hinyokika Kiyo 52:79
44	2006	36	M	Flank pain	R	Kidney	NA	Nephrectomy	BEN	NA	NA	NA	Alvarez Mugica M et al., Arch Esp Urol 59:195

45	2007	26	M	Incidental	R	Kidney	7	Nephrectomy	BEN	6	NED	POS	Constantinidis C et al., The Can J Urol 14:3583
46	2007	70	M	Flank pain and macrohematuria	L	Kidney	15	Nephrectomy	BEN	6	NED	POS	Znati K et al., Reviews in Urol 9:36
47	2007	51	F	Flank pain	L	Kidney	4	Nephrectomy	BEN	10	NED	POS	Bozkurt SU et al., APMIS 115:259
48	2007	66	F	Abdominal mass and macrohematuria	R	Kidney	11	Nephrectomy	BEN	NA	NA	NA	Kakoi N et al., Japn J Urol Surg 20 supple 598
49	2007	60s	M	Incidental	R	Kidney	3	Nephrectomy	BEN	3	NED	NA	Yoshida T et al., Hinyokika Kiyo 53:745
50	2008	34	F	Flank pain	L	Kidney	9	Nephrectomy	MAL	21	NED	POS	Magro G et al., APMIS 115:1020
51	2008	67	M	Macrohematuria	L	Kidney	7	Nephrectomy	BEN	10	NED	POS	Amano T et al., Hinyokika Kiyo 54:357
52	2008	44	F	Incidental	L	Kidney	5.8	Nephrectomy	BEN	40	NED	POS	Hirabayashi J et al., Hinyokika Kiyo 54:357
53	2009	75	F	Incidental	L	Kidney	4.5	Nephrectomy	BEN	9	NED	POS	Hirano D et al., Mod Mol Morphol 42:239
54	2009	64	F	Cough	L	Kidney	2.5	Biopsy	BEN	12	NED	POS	Petrella F et al., Minerca Chir 64:669
55	2009	35	M	Incidental	R	Kidney	8	Partial nephrectomy	BEN	NA	NA	POS	Makris A et al., Can J Urol 16:4854
56	2009	72	F	Abdominal mass	L	Kidney	19	Nephrectomy	MAL	NA	NA	NA	Marzi M et al., Urologia 76:112
57	2009	76	F	Incidental	R	Kidney	2.5	Nephrectomy	BEN	48	NED	POS	Yoneyama T et al., Hinyokika Kiyo 55:479
58	2009	50	M	Incidental	L	Kidney	5.5	Nephrectomy	BEN	NA	NED	POS	Matsumoto T et al., Japn J Urol Surg 22:230
59	2009	63	M	Incidental	L	Kidney	5.3	Nephrectomy	MAL	NA	NA	POS	Murayama S et al., Japn J Urol Surg 22:230
60	2009	51	F	Incidental	R	Kidney	12	Nephrectomy	BEN	NA	NA	POS	Ogushi S et al., Japn J Urol Surg 22:230
61	2009	75	M	NA	L	Kidney	3	Nephroureterec-tomy	BEN	NA	NA	POS	Kobori Y et al., Hinyokika Kiyo 55:305
62	2010	39	M	Dysuria	L	Kidney	25	Nephrectomy	BEN	12	NED	POS	Taza L et al., Actas Urol Esp 34:568
63	2010	39	F	Abdominal fullness	L	Kidney	20	Embolization and nephrectomy	BEN	6	NED	POS	Yamaguchi Y et al., Hinyokika Kiyo 56:435
64	2011	44	M	Macrohematuria	L	Kidney	NA	Embolization and nephrectomy	BEN	NA	NA	NA	Saegusa M et al., Nishinihon J Urol 68:187
65	2011	52	F	Abdominal pain	R	Kidney	18	Nephrectomy and thrombectomy	BEN	6	NED	POS	Naveen HN et al., Urol Ann 3:158
66	2011	72	F	Abdominal mass	L	Kidney	19	Nephrectomy	MAL	15	NED	POS (focal)	Marzi M et al., Minerva Urol Nephrol 63:109
67	2011	50	F	Flank pain	R	Kidney	15	Nephrectomy	MAL	30	NED	POS	Tsan-Yu Hsieh, Diag Pathol 6:96
Our case		48	M	Abdominal mass	R	Kidney	29	Nephrectomy	BEN	107	NED	POS (55%)	

M = Male; F = female; NA = not available; R = right; L = left; BE = benign; MAL = malignant; DNOD = died not of disease; NED = no evidence of disease; POS = positive.
* CD34 immunoreactivity (the extent of positive area is shown in parentheses, if information is available).

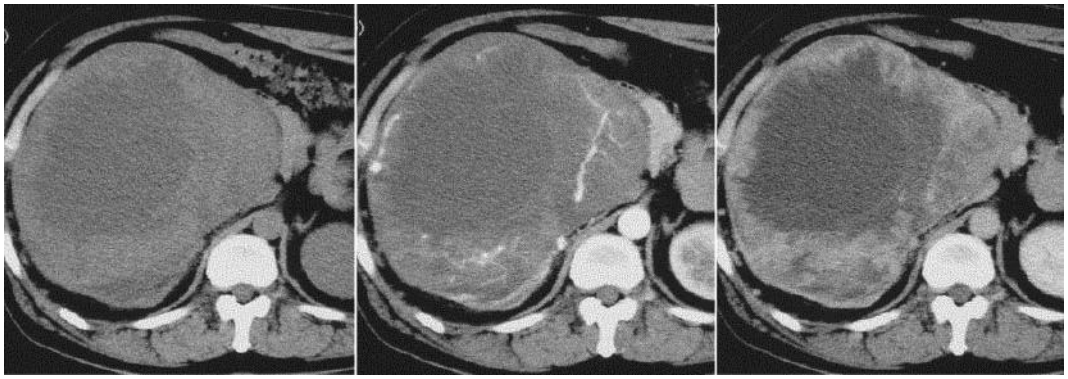

Fig. 1. CT findings. A large right renal tumor was observed on plain CT (left). Blood vessels were enhanced in the early phase (middle), and the tumor was enhanced in the delayed phase (right).

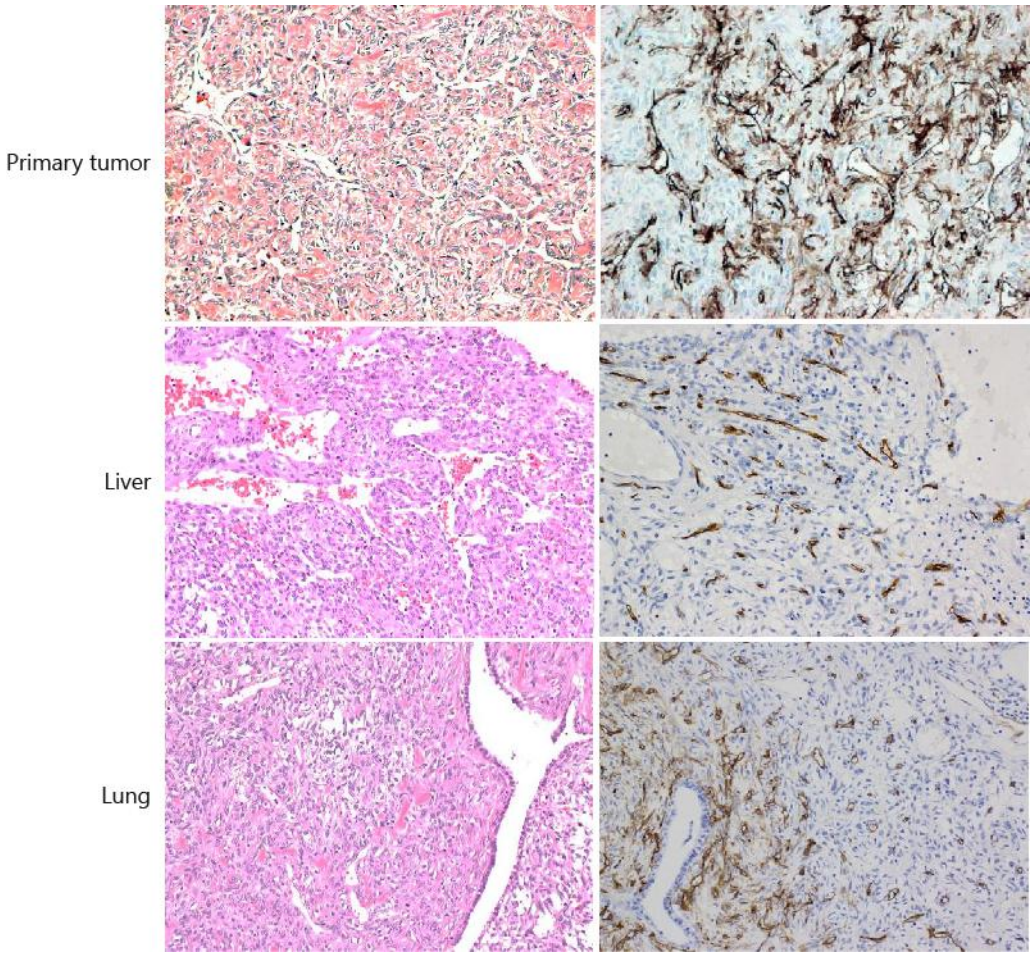

Fig. 2. Microscopic features of the solitary fibrous tumor. The primary renal tumor displayed a hemangiopericytomatous growth pattern (upper left). Immunohistochemical staining of CD34 was positive in 55% of the primary tumor (upper right). Resected tissue from the liver (middle left) and lungs (lower left). CD34 expression was completely absent from the resected liver tumor (middle right) but weakly positive in the resected lung tumor (lower right).

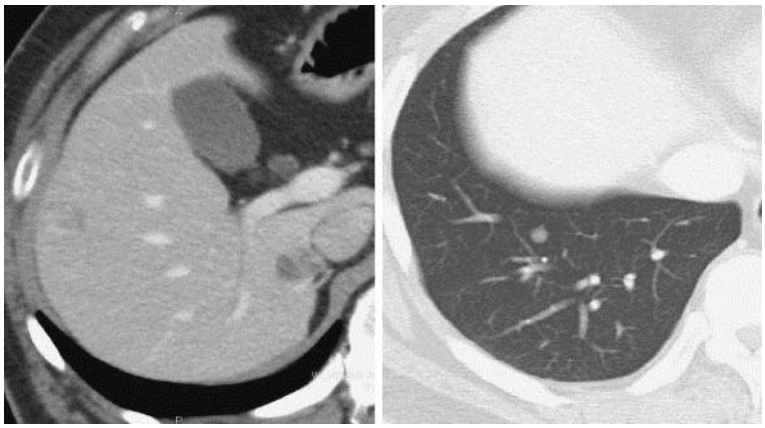

Fig. 3. Follow-up computed tomography revealed liver metastasis and lung metastasis.