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Case Report

A Woman With Persistent Abdominal Pain in the Setting of a Solitary Fibrous Adrenal Tumor

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ABSTRACT

Background/Objective: We present a case of a woman with persistent abdominal pain, leading to the discovery of a rare solitary fibrous tumor (SFT) in the adrenal gland. The rarity of adrenal SFTs and their diagnostic challenges make this case noteworthy. The objective of this report is to describe a patient with an adrenal SFT, highlighting the unique diagnostic approach and management considerations for this rare condition.

Case Report: A 35-year-old woman presented with chronic abdominal pain and was referred to the endocrinology clinic for evaluation of an adrenal incidentaloma. Computed tomography imaging revealed a 4 cm homogeneous right adrenal mass, with precontrast Hounsfield units of 1 and an absolute contrast washout of 60%. A prior computed tomography scan from 6 years earlier showed a 1.5 cm adenoma with similar characteristics. Despite normal hormonal levels, the tumor's growth and the patient's symptoms prompted surgical referral. The patient underwent robotic-assisted laparoscopic right adrenalectomy. Pathological examination identified a well-circumscribed SFT, measuring 3.7 × 3.6 × 2.9 cm.

Discussion: Adrenal SFTs are rare, typically hormonally inactive, well-circumscribed masses that often present with abdominal pain. While more common in the pleura, adrenal SFTs can mimic other benign adrenal lesions on imaging, complicating diagnosis. Histopathology is essential for accurate diagnosis, and surgical resection remains the main treatment.

Conclusion: This case highlights the diagnostic challenges of adrenal SFTs, which can mimic other benign lesions. Despite significant growth, the tumor was histologically benign with low malignancy risk. Clinicians should consider adrenal SFTs in the differential diagnosis of incidental adrenal masses with atypical imaging features.

Introduction

Solitary fibrous tumors (SFTs) were first described as originating from pleural tissue in the thoracic cavity[1] but have since been reported in various extra-pleural locations, including the retroperitoneum and adrenal glands.[2] Adrenal SFTs are considered rare, with only approximately 21 cases reported as of 2022.[3] The majority of these cases present as hormonally inactive, benign lesions with a low risk for malignancy, although there is significant variation in tumor size and patient demographics. SFTs in the adrenal glands tend to present in a wide age range, with a slight male preponderance.[3] When located in the retroperitoneum, these tumors may cause symptoms, typically due to their size and the compressive effects on surrounding structures, with abdominal pain being a common presenting symptom. The substantial size of the tumor often serves as an indication for surgical removal. In addition, few cases have been associated with fever of unknown origin, which were later identified as adrenal solitary fibrous tumors.[4,5]

Abbreviations: CT, computed tomography; HU, Hounsfield unit; SFT, solitary fibrous tumor.

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Macroscopically, adrenal SFTs are usually well-circumscribed, with a gray-white to yellow-white color, and may vary in size. Microscopically, they are characterized by a patternless cell architecture, the presence of spindle-shaped cells, and staghorn-like blood vessels.[2] Despite the rarity of adrenal SFTs, they are important to recognize due to their potential for significant growth and the clinical challenges they pose in diagnosis and management. Medical societies currently rely on case reports to increase awareness and provide insight into the presentation, diagnosis, and management of these tumors.

This report presents a 35-year-old woman with chronic abdominal pain, leading to the discovery of a 4 cm adrenal mass. Despite benign imaging and biochemical findings, significant tumor growth over 6 years required surgical removal, and histopathology confirmed an adrenal solitary fibrous tumor. This case underscores the diagnostic challenges of adrenal solitary fibrous tumors, which can mimic other benign lesions and present with symptoms, such as abdominal pain. While adrenal SFTs are rare, the patient's history of abdominal pain, in the context of significant tumor growth, offers a valuable contribution to the literature by highlighting a potential clinical feature that may warrant greater attention in future cases.

Highlights

- Adrenal solitary fibrous tumors are rare nonfunctioning tumors and generally benign
- Histologically, adrenal SFTs show spindle cells in a patternless arrangement
- Immunohistochemistry is typically positive for CD34 and STAT6
- Adrenal SFTs may grow significantly despite being hormonally inactive
- Long-term follow-up is crucial due to the potential for recurrence

Clinical Relevance

Due to the rarity of adrenal solitary fibrous tumors, there is no consensus on optimal management. This case highlights their potential for significant growth, characteristic histology, and benign nature, contributing to a better understanding of their clinical behavior and overall risk and management.

Case Report

A 35-year-old woman presented to the endocrinology clinic with a chief complaint of chronic abdominal pain. The patient described her abdominal pain as persistent and moderate for several months and was not associated with any other gastrointestinal symptoms such as nausea, vomiting, or changes in bowel movements. She had no significant medical or familial history of adrenal disorders.

Upon physical examination, the patient appeared well-nourished and in no acute distress. Abdominal examination revealed mild tenderness upon palpation in the right upper quadrant without palpable masses or signs of peritoneal irritation. No hepatosplenomegaly was noted, and there were no palpable lymph nodes. The remainder of the physical exam was unremarkable.

During evaluation, a computed tomography (CT) scan of her abdomen and pelvis, both with and without intravenous contrast, revealed a homogeneous 4 cm mass in the right adrenal gland (Fig. 1). The precontrast Hounsfield unit (HU) value was 1, with an absolute contrast washout of 60%. A comparison with a CT scan from 6 years prior showed a similar lesion, although smaller at 1.5 cm (Fig. 1).

Biochemical tests were performed to assess for hormonal hypersecretion. The results were all within normal reference ranges (Table). Specifically, the morning cortisol after 1 mg dexamethasone suppression was measured at 1.0 mcg/dL (reference range: < 1.8 mcg/dL). Serum aldosterone levels were 1.8 ng/dL (reference range: 0-30 ng/dL), and serum renin was 1.363 ng/mL/h (reference range: 0.167-5.380 ng/mL/h). Additionally, serum metanephrines were found to be < 10.0 pg/mL (reference range: 0.0-88.0 pg/mL), and serum normetanephrines were 46.6 pg/mL (reference range: 0.0-210 pg/mL). Given the patient's persistent abdominal pain and the growth of the lesion, she was referred for surgical intervention.

The patient subsequently underwent a robotic-assisted laparoscopic right adrenalectomy. Pathological examination revealed a well-circumscribed, fleshy tumor measuring 3.7 × 3.6 × 2.9 cm. Microscopic analysis revealed spindle cells arranged in a disordered pattern with hyalinized staghorn-like blood vessels with low mitotic activity of 3/10 high-power fields (hpf). Immunohistochemical analysis showed positive staining for CD34 and STAT6 (Fig. 2).

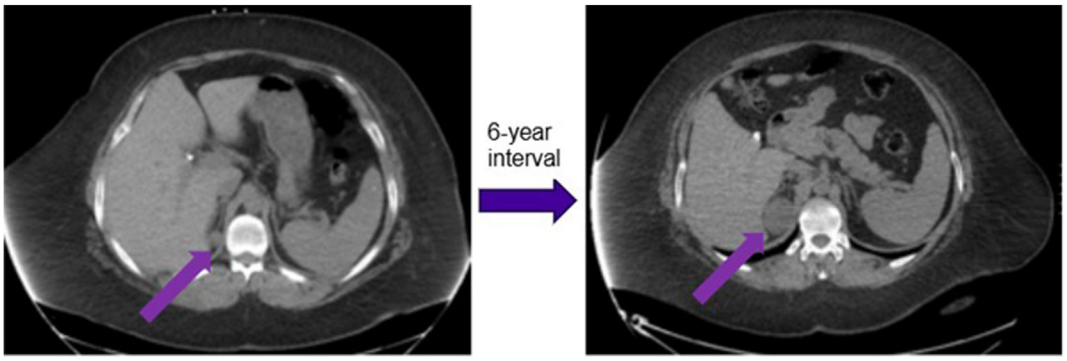

Fig. 1. Radiographic CT imaging of a solitary fibrous tumor of the right adrenal gland over a six-year span. *CT* = computed tomography.

Table
Hormonal Investigation to Assess for Functionality of Adrenal Tumor

Hormonal workup	Results	Reference range
Morning cortisol after 1 mg dexamethasone suppression	1.0 mcg/dL	<1.8 mcg/dL
Serum aldosterone	1.8 ng/dL	0-30 ng/dL
Serum renin	1.363 ng/mL/h	0.167-5.380 ng/mL/h
Serum metanephrines	<10.0 pg/mL	0.0-88.0 pg/mL
Serum normetanephrines	46.6 pg/mL	0.0-210 pg/mL

Results demonstrate hormonally nonfunctioning tumor.

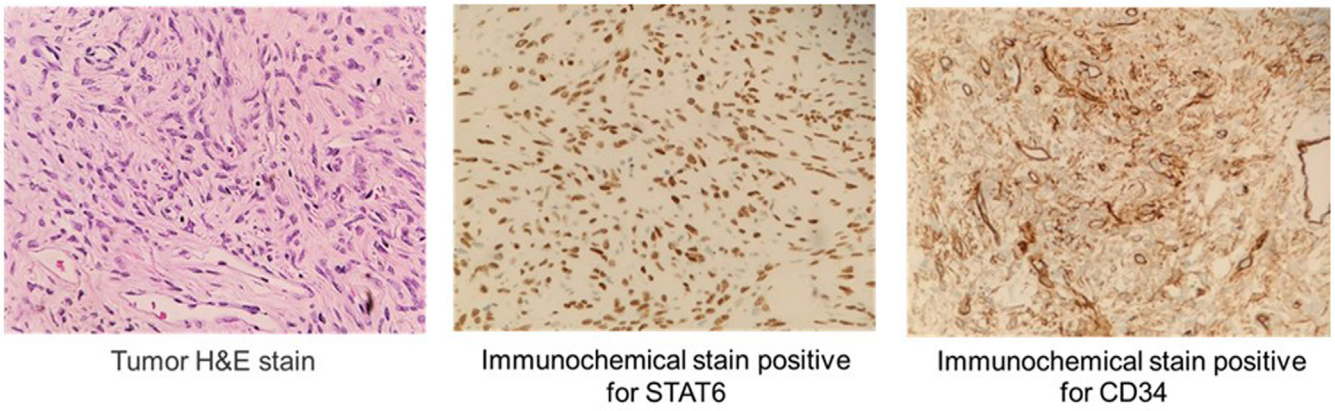

Fig. 2. Microscopic histology of a solitary fibrous tumor of the adrenal gland comprised of spindle cells showing mild nuclear enlargement arranged in a haphazard fashion. Hyalinized staghorn-like blood vessels are identified in the background and low mitotic activity of 3/10 hpf. *H&E* = hematoxylin and eosin; *hpf* = high-power field.

The patient's immediate postoperative course was uneventful. However, approximately 10 days postsurgery, the patient presented to the emergency department with acute right-sided abdominal pain. Abdominal CT imaging confirmed the removal of the adrenal tumor but revealed an acute hematoma in the liver. No surgical intervention was required, and the patient was discharged home for clinical monitoring. At her 6-month follow-up, she was asymptomatic, and imaging showed no evidence of tumor recurrence.

Discussion

This case describes a 35-year-old woman with chronic abdominal pain, ultimately diagnosed with a hormonally inactive adrenal SFT of the right adrenal gland. Over a span of 6 years, the tumor grew by 2.5 cm, from 1.5 cm to 4 cm, with a growth rate of approximately 4.17 mm per year. This rate exceeds the typical growth observed in nonfunctioning adrenal lesions, which increase by about 2 mm over a median of 52.8 months.[6,7] Although adrenal SFTs are rare, they have been reported to range in size from 2.5 cm to 20 cm, with varying growth rates.[2,8,9] In this case, the tumor's substantial growth over 6 years resulted in the patient's symptoms of chronic abdominal pain. However, unlike a series of 9 patients with abdominal pain, all of whom had larger tumors,[3] the patient's 4 cm tumor suggests that even smaller adrenal SFTs can cause symptoms typically seen with larger masses.

The HU values for adrenal SFTs on CT scans can vary based on tumor composition, such as fibrous tissue content and vascularity. Although specific HU values for adrenal SFTs are not well-defined, literature suggests precontrast HU values for retroperitoneal SFTs typically range from 20 to 50 HU.[10] In contrast, the HU of 1 in this case report falls outside this typical range but remains within the realm of possibility, especially for a fibrous tumor with limited vascularity.

The patient's biochemical workup, including measurements of cortisol, aldosterone, and metanephrines, showed no evidence of hormonal hypersecretion, confirming the nonfunctional nature of the tumor. These findings are consistent with most cases of adrenal SFT, where hormonal overproduction is typically absent.[2,3] Surgical resection remains the primary treatment for adrenal SFTs, with chemotherapy and radiotherapy rarely indicated.[11] Histopathological examination, with the use of specific immunohistochemical stains, is essential for diagnosis.[12] In this case, the tumor was positive for CD34 and STAT6, consistent with the diagnosis of SFT and helping to exclude other adrenal lesions. These findings align with existing literature, where adrenal SFTs are typically immunoreactive for CD34 and STAT6, while negative for desmin, SMA, SOX10, and S100 proteins.[13] Differential diagnosis of adrenal SFT must also consider a range of other conditions, including the Ewing family of tumors, Wilms tumor, desmoplastic small round cell tumor, neuroblastoma, small cell carcinoma, hematolymphoid neoplasms, round cell sarcoma, Merkel cell carcinoma, and embryonal rhabdomyosarcoma.[3] Crucially, these tumors do not express CD34 and STAT6, which helps differentiate them from adrenal SFT.[13]

Histologically, adrenal SFTs demonstrate spindle cell morphology, patternless proliferation, and staghorn-like blood vessels,[2] characteristics also present in this case. Mitotic activity was low, with a mitotic count of 3/10 hpf, which, along with the absence of necrosis, placed this patient's tumor in the low-risk category for malignancy. This histological profile is consistent with the typical features of adrenal SFTs, which generally exhibit low mitotic activity and low-to-moderate nuclear atypia.[2,3,12,14]

While most known adrenal SFTs are benign,[3] long-term follow-up after surgical resection is recommended due to documented cases of tumor recurrence over time.[3,15] Approximately 10% to 15% of SFTs show recurrent and/or metastatic disease.[16] Risk stratification models, such as the one by Demicco et al (Fig. 3), assess

Risk Factor	Cut-Off	Points Assigned	
		3-Variable Model	4-Variable Model
Patient age (years)	<55	0	0
	>55	1	1
Mitoses/mm^2	0	0	0
	0.5–1.5	1	1
	≥2	2	2
Tumor size (cm)	0–4.9	0	0
	5–9.9	1	1
	10–14.9	2	2
	≥15	3	3
Tumor necrosis	<10%	N/A	0
	≥10%	N/A	1
Risk	Low	0–2 points	0–3 points
	Intermediate	3–4 points	4–5 points
	High	5–6 points	6–7 points

Fig. 3. Risk stratification model for solitary fibrous tumors proposed by Demicco et al 2017.

malignant potential in solitary fibrous tumors using variables like age, tumor size, mitotic count, and necrosis to guide management.[16,17] In the 3-variable model (excluding necrosis), scores of 0-2 indicate low risk, 3-4 intermediate risk, and 5-6 high risk for malignancy. The 4-variable model adjusts these ranges, with low risk defined as 0-3, intermediate risk as 4-5, and high risk as 6-7. Based on this model, 45% of patients were low risk, 37% intermediate risk, and 18% high risk, with the latter group showing 100% metastatic risk by 5 years.[17] Regardless of risk, surgical resection remains the primary treatment for SFTs, with histological examination and immunohistochemical staining being crucial for diagnosis.[11,12]

Adrenal SFTs present diagnostic challenges, particularly in smaller tumors that can cause abdominal pain symptoms. Despite being hormonally nonfunctional, the tumor's growth highlights the need for careful monitoring and long-term follow-up. Histopathological and immunohistochemical analysis, including CD34 and STAT6 positivity, are essential for diagnosis. While most adrenal SFTs are benign and low-risk, surgical resection is crucial, and this case adds to the literature by emphasizing the importance of considering SFTs in the differential diagnosis of nonfunctional adrenal masses.

This case report highlights solitary fibrous tumors as a potential consideration for adrenal nonfunctioning tumors, particularly when imaging findings are atypical. Recognizing the potential for substantial growth over time in these tumors is crucial, as it may warrant surgical intervention and long-term postoperative monitoring to clinically assess tumor recurrence. Continued case reporting may refine diagnostic strategies and management approaches for these rare tumors.

Conclusion

Adrenal SFTs are rare, nonfunctional lesions that can present with symptoms, such as chronic abdominal pain, despite their benign nature. This case highlights the importance of considering SFTs in the differential diagnosis of adrenal incidentalomas, especially when there is substantial growth or unusual imaging features. Early diagnosis and surgical resection remain the primary treatment, with histopathological and immunohistochemical analysis essential for confirming the diagnosis. Due to the potential for recurrence, long-term follow-up is recommended. Continued documentation and analysis of these rare tumors will further enhance diagnostic accuracy and guide therapeutic strategies.

Patient consent

Manuscript acknowledges that patient consent for case report publication was obtained.

Disclosure

The authors have no conflicts of interest to disclose.

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