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Case Report

Bilateral Adrenal Leiomyoma Presenting as Progressive Non-Functioning Adrenal Mass in a Young Female: A Rare Case Report

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Abstract

Background:

Adrenal masses are increasingly detected with modern imaging, but adrenal leiomyomas (LMs) are exceptionally rare benign mesenchymal tumors. Bilateral involvement is particularly uncommon and often difficult to diagnose preoperatively.

Case presentation:

A 30-year-old female with celiac disease and chronic anemia presented with intermittent left flank pain. She had previously undergone right adrenalectomy for a lesion initially diagnosed as ganglioneuroma. Imaging revealed a progressively enlarging nonfunctioning left adrenal mass with atypical features. Hormonal evaluation was negative. Laparoscopic left adrenalectomy was performed, and histopathology confirmed bilateral adrenal leiomyoma.

Conclusion:

This case highlights the diagnostic dilemma of adrenal tumors, especially adrenal LMs and supports surgical excision for symptomatic or enlarging nonfunctioning adrenal masses.

Introduction

Adrenal masses are relatively uncommon, with a reported prevalence ranging from approximately 0.2% in younger individuals to up to 7% in older populations [1]. With the widespread use of modern cross-sectional imaging, adrenal tumors are now detected incidentally in approximately 4 to 8 percent of patients, often during evaluation for unrelated conditions. These lesions may arise from either the adrenal cortex or medulla and most commonly include adenomas, pheochromocytomas, and adrenocortical carcinomas. Less frequently, adrenal masses represent metastatic disease or originate from mesenchymal or connective tissue elements within the adrenal gland [2- 4].

Among the rarest of these tumors are adrenal leiomyomas or LMs, which are benign mesenchymal neoplasms derived from smooth muscle cells. To date, only 28 cases have been reported in the English literature, with a median age at diagnosis of 34.5 years and a clear female predominance. A substantial proportion of reported cases have occurred in patients with immunodeficiency, particularly those with HIV infection, suggesting a possible association. Even more exceptional are bilateral adrenal LMs, for which the true prevalence remains unknown due to the extreme rarity of reported cases [5,6].

Clinically, adrenal LMs may remain asymptomatic for prolonged periods, particularly when small. Larger tumors may become symptomatic, most commonly presenting with dull abdominal or flank pain related to

mass effect. As a result, these tumors are frequently discovered incidentally on imaging and may closely mimic malignant adrenal lesions, often prompting surgical intervention to establish a definitive diagnosis [7].

Here, we report the case of a 30-year-old female with celiac disease and chronic anemia who presented with a progressively enlarging left adrenal mass associated with intermittent flank pain, ultimately diagnosed as bilateral adrenal LM.

Case Report

A 30-year-old female with a known history of celiac disease and chronic iron deficiency anemia was referred to the Endocrine Surgery clinic following detection of a left adrenal mass on computed tomography. Her surgical history was significant for a open right adrenalectomy performed in 2017 at an outside institution for a right adrenal mass, which was initially reported on histopathology as a ganglioneuroma. At presentation, the patient reported intermittent dull pain in the left upper abdomen and flank, occasionally accompanied by nausea without vomiting. She denied fever, unintentional weight loss, or other systemic symptoms. There was no family history of malignancy. Physical examination revealed no visible or palpable abdominal masses. The abdomen was soft, non tender, and non distended, with no organomegaly.

Preoperative laboratory evaluation demonstrated normal serum electrolytes, with sodium of 138 mmol/L and potassium of 4.8 mmol/L. Baseline serum cortisol was 336 nmol/L and showed appropriate suppression to less than 28 nmol/L following a low dose dexamethasone suppression test, excluding autonomous cortisol secretion. Plasma and urinary metanephrines were within normal limits, effectively ruling out a catecholamine secreting tumor. Renal function was preserved. Thyroid function testing showed a mildly elevated thyroid stimulating hormone level of 5.2 mIU/L with a normal free thyroxine level of 12 pmol/L. Hematological evaluation confirmed chronic iron deficiency anemia, with a hemoglobin level of 9g/dL and low serum iron of 4 micromol/L. Serum vitamin D3 was 114 nmol/L , and HIV screening was negative. Bone mineral density assessment using dual energy x ray absorptiometry demonstrated osteoporosis, with a lumbar spine T score of -2.8 and a femoral neck T score of -1.8.

A contrast enhanced computed tomography scan of the abdomen and pelvis using an adrenal protocol was performed in October 2023 and compared with imaging from October 2021. This demonstrated interval progression of the left adrenal lesion. The left adrenal gland was largely replaced by a complex heterogeneous mass measuring approximately 5.0 by 4.5 by 5.7 centimeters, increased from 4 centimeters on prior imaging. The lesion exerted mass effect on the upper pole of the left kidney, contained internal calcifications, and demonstrated progressive enhancement on portal venous and delayed phases without washout. A new 1.5 centimeter exophytic nodule extending into the adrenal splenic space was also identified, showing peripheral enhancement with central fluid or necrotic change. Multiple small mesenteric and bilateral inguinal lymph

nodes were noted and considered nonspecific, likely related to the patient's underlying celiac disease.

Despite a negative hormonal workup and initial consideration of a nonfunctioning adrenal adenoma, the patient's persistent symptoms, progressive tumor enlargement, and atypical imaging features supported surgical intervention. She subsequently underwent laparoscopic left adrenalectomy. Intraoperative findings were consistent with a large left adrenal tumor, correlating with preoperative imaging.

Histopathological examination revealed a benign spindle cell neoplasm consistent with adrenal LM, with clear surgical margins. Review of the original histopathological slides from the prior right adrenalectomy reclassified the lesion as a benign adrenal LM with focal degenerative changes, confirming the diagnosis of bilateral adrenal LM.

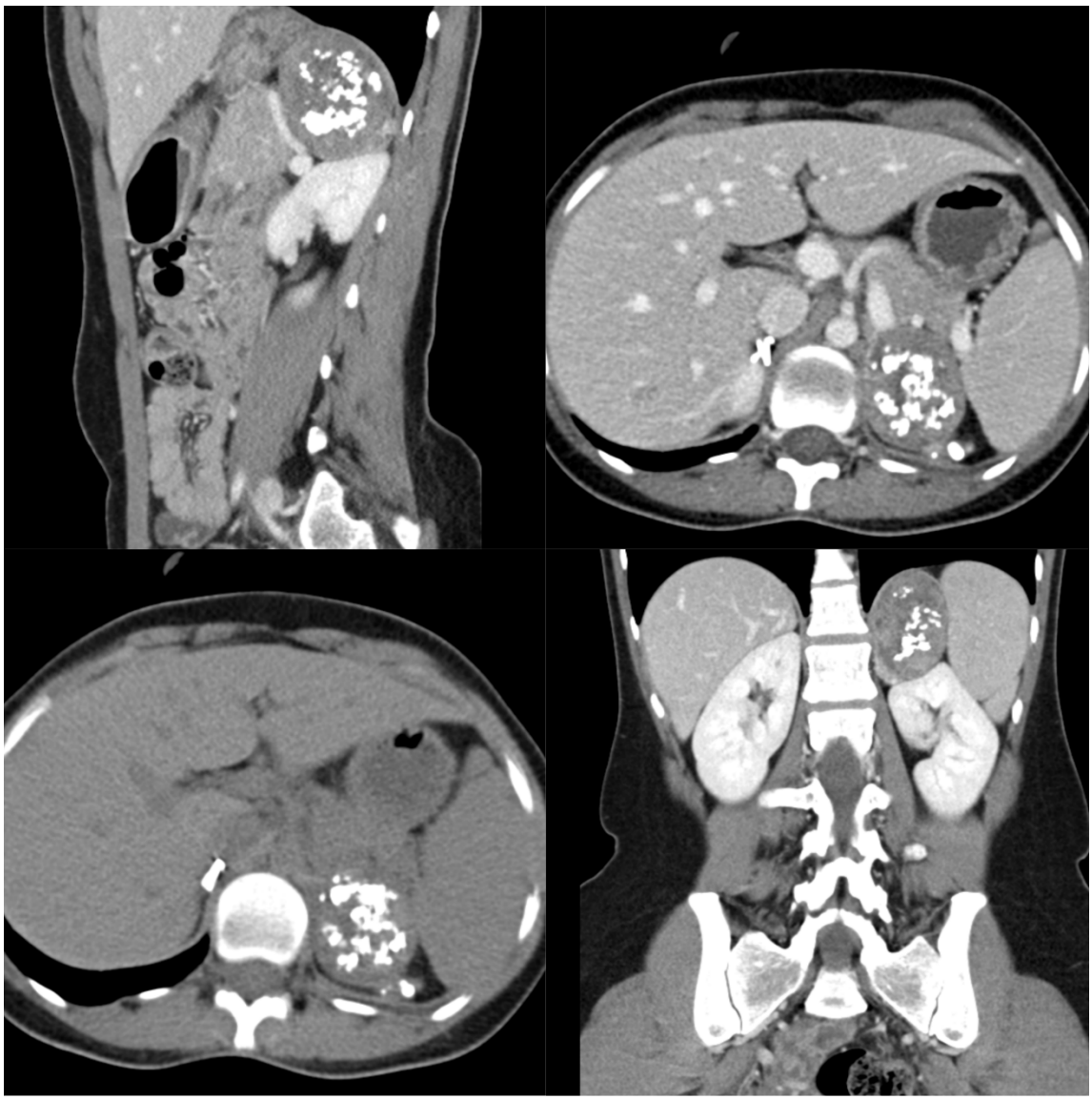

Figure 1. Contrast-enhanced CT scan demonstrating a large heterogeneous left adrenal mass with internal calcifications and a new exophytic component

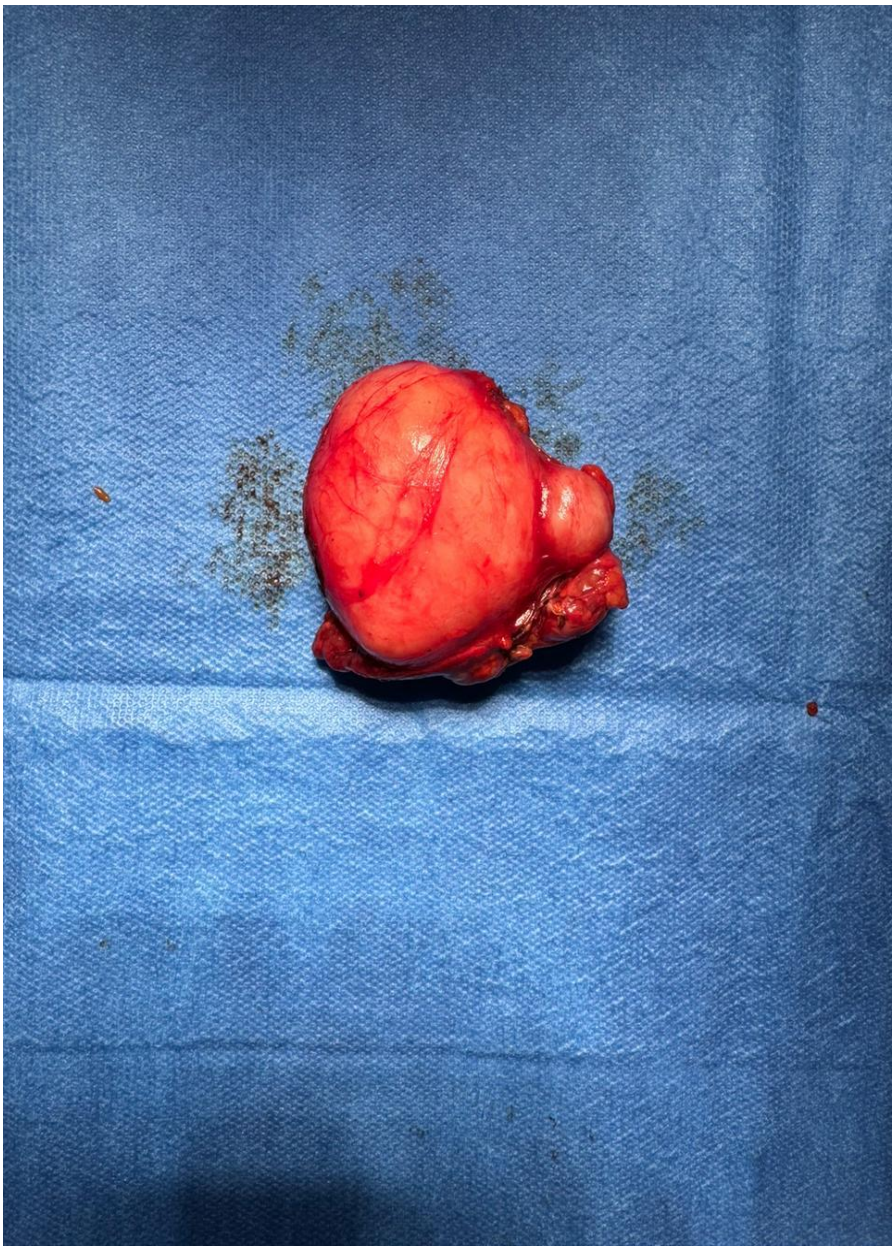

Figure 2. Gross specimen of the resected left adrenal mass following laparoscopic adrenalectomy, showing a well-circumscribed solid tumor consistent with adrenal leiomyoma

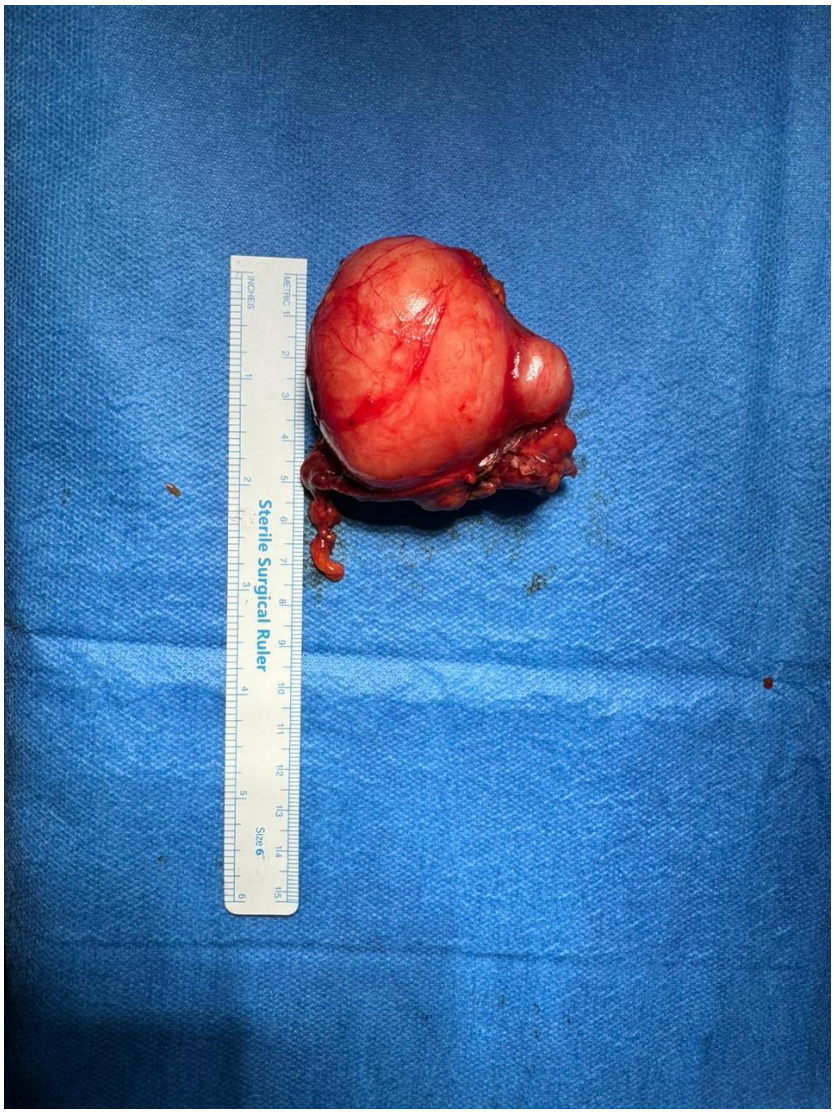

Figure 3. Gross specimen of the resected left adrenal leiomyoma shown in comparison with a reference scale, demonstrating the size of the tumor following excision

Discussion

Adrenal LMs are exceptionally rare benign mesenchymal tumors arising from smooth muscle cells. In contrast, the majority of adrenal incidentalomas are far more common epithelial or neuroendocrine lesions. Approximately 75% of adrenal incidentalomas are nonfunctioning adenomas, while 10 to 12% secrete cortisol, 7 to 10% represent pheochromocytomas, 2.5-6% aldosteronomas, 8 to 11% adrenocortical carcinomas, and 5 to 7% metastatic lesions. Within this context, adrenal LMs represent a significant diagnostic challenge due to their extreme rarity, nonspecific imaging characteristics, and lack of hormonal activity [2].

The rarity of adrenal LMs has been consistently emphasized in the literature. A systematic review by Sakellariou et al identified only a limited number of reported cases worldwide, highlighting that adrenal LMs constitute a very small fraction of adrenal tumors and are predominantly described as isolated case reports or

small series [4]. Most lesions were nonfunctioning, incidentally detected, and unilateral. While the present case aligns with the reported demographic trend of young to middle aged females, it is distinguished by true bilateral involvement, a finding reported only exceptionally [2,6,7]. Bilateral adrenal LMs have been described only sporadically, including isolated pediatric and adult cases, underscoring the absence of standardized management strategies for this presentation [8,9].

Bilateral adrenal LMs present a distinct diagnostic challenge. Bilateral adrenal masses are more commonly associated with metastatic disease, congenital adrenal hyperplasia, infiltrative disorders, or bilateral pheochromocytomas, making benign mesenchymal tumors an unlikely preoperative diagnosis. Published bilateral cases consistently report diagnostic uncertainty and concern for malignancy prior to histological confirmation [2,7]. In this patient, the metachronous presentation further complicated recognition and highlights the importance of long term surveillance in patients with previously resected rare adrenal tumors.

An association between adrenal LMs and immunodeficiency, particularly HIV infection and Epstein Barr virus related smooth muscle tumors, has been proposed in earlier reports [4,5]. However, increasing evidence demonstrates that adrenal LMs also occur in immunocompetent individuals, including pediatric patients and young adults [6,7]. The absence of immunodeficiency in this case supports the view that immune compromise is not a prerequisite for tumor development.

Clinically, adrenal LMs are typically indolent. When symptoms occur, they result from mass effect rather than hormonal activity. Most patients present with vague abdominal or flank pain, while less common manifestations such as lower extremity edema or spider angiomas have been reported in cases involving invasion of the inferior vena cava [2,4,5]. In the present case, progressive tumor growth correlated with symptom development, supporting surgical intervention despite biochemical inactivity and reinforcing the importance of symptom burden and growth dynamics in adrenal mass management.

Radiologically, adrenal LMs lack distinguishing imaging features. Large size, heterogeneous enhancement, internal calcifications, and absence of contrast washout significantly overlap with imaging characteristics of malignant adrenal neoplasms [3,4,5,7]. In this case, interval growth, atypical enhancement, and the development of an exophytic component raised concern for malignancy, emphasizing that imaging alone is insufficient to reliably distinguish adrenal LMs from aggressive adrenal tumors.

Biochemically, adrenal LMs are almost uniformly nonfunctioning. Comprehensive endocrine evaluation remains essential to exclude hormonally active lesions in accordance with established guidelines [1]. However, hormonal silence does not guarantee benign behavior on imaging, and reliance on biochemical testing alone may delay definitive management.

Histopathological examination remains the cornerstone of diagnosis. Histological and immunohistochemical evaluation confirms smooth muscle differentiation and excludes malignant features [4,5]. Complete surgical excision is curative, with no reported cases of malignant transformation or recurrence. Importantly, expert pathological review is essential, as illustrated by reclassification of the patient's prior right adrenal lesion, highlighting the potential for misdiagnosis in rare adrenal tumors.

Conclusion

This case adds evidence to the limited literature on adrenal LMs by documenting a rare bilateral metachronous presentation in an immunocompetent young female. It underscores the diagnostic limitations of imaging and biochemical evaluation, reinforces the role of surgical excision in symptomatic or enlarging nonfunctioning adrenal masses, and highlights the importance of expert histopathological assessment in establishing an accurate diagnosis.

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