CASE REPORT OPEN ACCESS

Spontaneous Rupture of Adrenal Myelolipoma: A Case Report With Review of the Literature

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Received: 23 November 2024 | **Revised:** 26 February 2025 | **Accepted:** 14 March 2025

Keywords: adrenal gland | myelolipoma | spontaneous rupture

ABSTRACT

Introduction: Adrenal myelolipoma is often asymptomatic and is commonly diagnosed using imaging modalities. Herein, we report a case of adrenal myelolipoma detected through spontaneous rupture and review similar cases from the literature.

Case Presentation: A 27-year-old man was admitted to our hospital with sudden right flank pain. Abdominal computed tomography revealed right retroperitoneal hemorrhage due to a spontaneous rupture of a retroperitoneal tumor. He underwent emergency transcatheter arterial embolization because of his severe symptoms and poor general condition, which included abdominal pain and progressive anemia. We performed laparoscopic resection of the tumor with the right adrenal gland 3 months after the emergency embolization. Consequently, a definitive diagnosis of adrenal myelolipoma was confirmed.

Conclusion: Adrenal myelolipoma, although scarcely reported, can rupture spontaneously and lead to severe consequences. Treatment options may include percutaneous arterial embolization and subsequent surgery.

1 | Introduction

Adrenal myelolipoma is a rare disease with a clinically benign nature. It comprises adipose tissue and mature hematopoietic tissue, resembling bone marrow [1]. Adrenal myelolipoma is mostly found incidentally; however, cases are occasionally diagnosed through tumor rupture and the subsequent complications associated with hemorrhagic shock [2]. The management and treatment of adrenal myelolipoma are controversial as the tumor is typically asymptomatic. However, although infrequently, spontaneous ruptures, which are proven to be fatal, can occur. Herein, we report a case of the spontaneous rupturing of adrenal myelolipoma and consider the management and treatment indications for this condition.

2 | Case Presentation

A 27-year-old man visited a primary care doctor with a sudden onset of right flank pain. The symptoms were similar to those of appendicitis, and the patient was treated with antibiotics and analgesics. Owing to persisting symptoms, he was admitted to our hospital for further evaluation. The enhanced abdominal computed tomography (CT) scan revealed a 12 × 11 × 11 cm right retroperitoneal mass with a fat density area and widespread hemorrhage inside the tumor, along with surrounding hematoma (Figure 1). Emergency treatment was initiated because of persistent abdominal pain and anemia (10.0 g/dL of hemoglobin count). The patient had progressive anemia, but the vital signs were relatively stable without the need for blood transfusions. After the diagnosis of probable rupture of the retroperitoneal renal angiomyolipoma was established, emergency angiography and transcatheter arterial embolization were performed. The angiogram revealed the middle adrenal artery as the feeding artery of the tumor (Figure 2), and the rupture of the right adrenal myelolipoma was confirmed. The embolization was effective and led to the stabilization of the patient's general condition and immediate relief of symptoms. Symptoms and laboratory findings related to the massive hemorrhage resolved, and the patient

Summary

- Adrenal myelolipoma is a primarily benign tumor that rarely ruptures during its clinical course. However, its rupture can lead to fatal complications caused by retroperitoneal hemorrhage.
- Therefore, it is crucial to include adrenal myelolipoma in the differential diagnosis of retroperitoneal hemorrhage to avoid potential mortality.

was discharged 2 weeks after embolization in stable general condition. The decision on the timing of surgery was controversial because of the unknown effects after a hemorrhagic event. It was considered that a period of time was needed for the general condition and the bleeding site to stabilize. We performed laparoscopic resection of the adrenal gland tumor 3 months after embolization (Figure 3). Because the tumor adhered to the surrounding tissue, careful separation was required during the surgical procedure. The surgery was completed in approximately 9 h with minimal operative bleeding. Surgical resection was followed by the pathological diagnosis of myelolipoma in the right adrenal gland. The postoperative course was uneventful, and no tumor recurrence was observed for several years postoperatively.

3 | Discussion

Adrenal myelolipoma is a rare disease in the surgical field, often diagnosed incidentally through medical checkups using diagnostic imaging such as ultrasonography. Autopsy findings suggest an incidence rate ranging from 0.08% to 0.4% [3]; however, with advancements in non-invasive enhanced imaging techniques, the prevalence has risen up to 10% [4]. In previous reports, incidental rupturing of adrenal myelolipoma with sudden onset and severe symptoms has been described. The spontaneous rupture can result in significant bleeding, leading to severe symptoms, and in some cases, result in retroperitoneal hemorrhage—a serious complication with potentially fatal consequences [5].

Ultrasonography, CT, and magnetic resonance imaging are useful techniques for the clinical diagnosis of adrenal myelolipoma, of which CT is the most effective and sensitive diagnosis, achieving a diagnostic accuracy of more than 90% [6]. Moreover, enhanced CT is beneficial for the accurate determination of bleeding grade and hemorrhagic site. In our case, emergency treatment was required because of severe flank pain and progressive anemia. Enhanced CT revealed a huge tumor with an area of fat density and widespread retroperitoneal hemorrhage; thus, immediate treatment for hemostasis was planned. After speculating the rupture of the renal angiomyolipoma in the probable differential diagnosis, we performed transcatheter arterial embolization. Angiography revealed that the tumor originated from the right adrenal gland because of the presence of the middle adrenal artery as a feeding artery. These examinations revealed that the clinical scenario was a spontaneous rupture of the adrenal myelolipoma. Therefore, renal angiomyolipoma, retroperitoneal lipoma, and liposarcoma should be considered in the differential diagnosis of retroperitoneal neoplasms, including fat-dense lesions. Furthermore, the occasional rupture of renal angiomyolipoma due to an intra-tumoral aneurysm can lead to retroperitoneal hemorrhage, making it a pertinent consideration [7]. To clarify the organ responsible for the hemorrhage, interventional radiology can play a crucial role in both the differential diagnosis and treatment of retroperitoneal tumors.

Previously, 22 cases of spontaneous rupture of adrenal myelolipomas have been reported in the literature [6, 8–18]. We have summarized the clinical features and management of these cases, including our experiences (Table 1). The disease occurs at

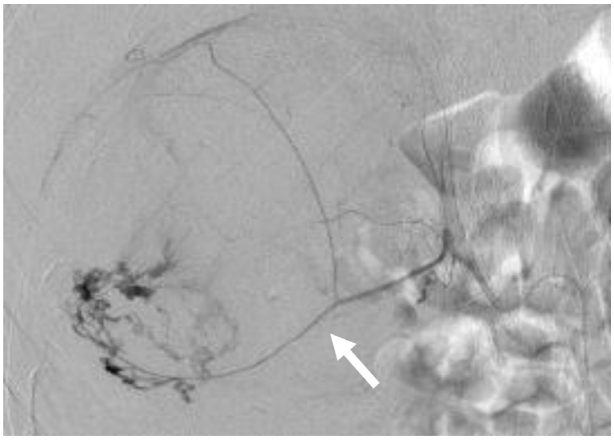

FIGURE 2 | Angiography showing bleeding from the middle adrenal artery (white arrow), suggesting that the tumor originates from the adrenal gland.

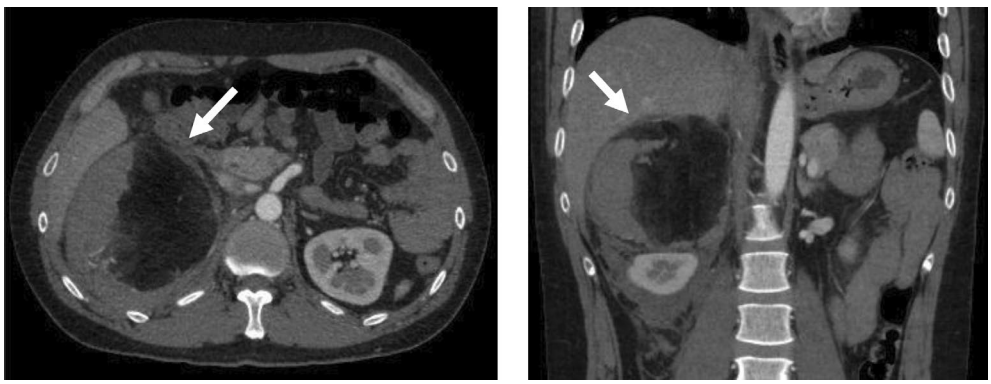

FIGURE 1 | Enhanced computed tomography showing a right retroperitoneal mass, including hemorrhage, surrounded by a hematoma (white arrows).

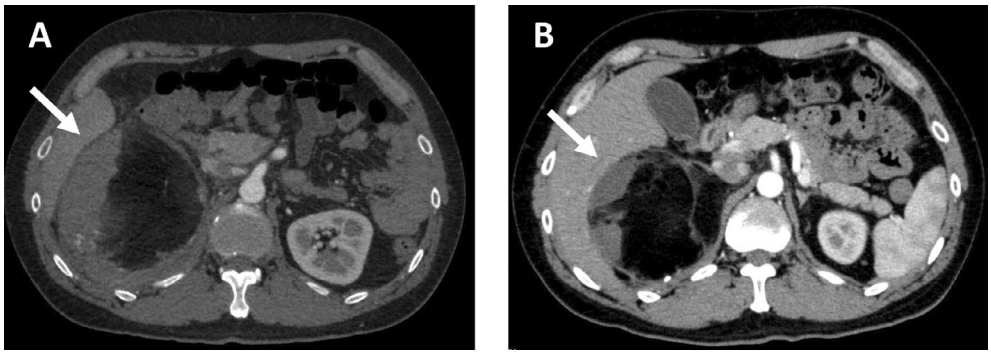

FIGURE 3 | The tumor shrank and the findings of tumor hemorrhage and surrounding inflammation were restored. (A) at onset prior to embolization, (B) 3 months after embolization.

TABLE 1 | Summary of 22 cases of ruptured adrenal myelolipoma.

	Number of cases (%)
Mean age (years, min–max)	46 (27–69)
Gender	
Male	19 (86%)
Female	3 (14%)
Laterality	
Right	17 (81%)
Left	5 (23%)
Mean tumor size (cm, min–max)	15 (7–35)
Diagnostic imaging	
CT only	12 (52%)
CT and angiography	7 (33%)
CT and ultrasonography	2 (10%)
CT and MRI	1 (5%)
Treatment	
Tumor resection with adrenalectomy	13 (57%)
Tumor resection with adrenalectomy and nephrectomy	6 (29%)
Tumor resection after TAE	3 (14%)

various ages and more frequently on the right side. CT has been the most useful modality for diagnosis; however, angiography has been useful in some cases, followed by transcatheter arterial embolization. These imaging examinations have revealed important findings for diagnosis and treatment strategies. In previous reports, all patients underwent successful surgical treatment under emergency or elective conditions. We believe that surgical treatment is essential for such patients. Notably, resection with nephrectomy was performed in several cases because of the difficulty in reaching a presurgical diagnosis and surgical adhesions. Therefore, we emphasize the significance of timely surgical intervention following enhanced CT and/or angiography to ensure relief from recurrent symptoms and to achieve a definitive diagnosis.

The surgical indication for adrenal myelolipoma is controversial because of its benign behavior. It has been reported that huge tumors larger than 6–10 cm should be operated on because of the potential for bleeding or symptoms [19, 20]. Large prospective studies of how larger adrenal myelolipomas are symptomatic or bleeding are needed to guide surgical treatment.

4 | Conclusions

Herein, we reported a case of spontaneous rupture of adrenal myelolipoma. Emergency angiography and transcatheter arterial embolization have enabled accurate diagnosis and treatment. Differential diagnosis and related treatment strategies should be considered in probable cases of rupture of adrenal myelolipomas.

Acknowledgments

The authors have nothing to report.

Ethics Statement

The authors have nothing to report.

Consent

An informed consent was obtained from the patient for the publication of this manuscript and the accompanying images.

Conflicts of Interest

The authors declare no conflicts of interest.

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