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Spontaneous Rupture of Adrenal Myelolipoma

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ABSTRACT

Adrenal myelolipoma is a benign tumor composed of adipose tissue and hematopoietic lesions. It is usually diagnosed incidentally in imaging tests such as computed tomogram (CT) or by patient symptoms such as retroperitoneal hemorrhage due to tumor rupture. We present a patient with retroperitoneal hemorrhage due to spontaneous rupture of a myelolipoma with enlargement of the mass and hemorrhage during the follow-up regimen of watchful waiting.

KEYWORDS: Rupture of Adrenal Myelolipoma; Enlargement of the mass and hemorrhage; Watchful waiting

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INTRODUCTION

Adrenal myelolipoma is a benign and endocrinologically non-functioning tumor generally composed of fat and hematopoietic elements. It is now found more frequently than in the past because of routine ultrasound and computerized tomography (CT) procedures. Rupture and bleeding of the myelolipoma is an infrequent complication resulting in a massive retroperitoneal hemorrhage. The spontaneous rupture of the myelolipoma that we report here is an unusual occurrence.

CASE REPORT

A 65-year-old man with a history of diabetes mellitus was admitted to the emergency department with persistent flank pain on the right side. He had no recent trauma, hematuria, voiding complaints, nausea/vomiting, or fever, and had tenderness and a palpable mass in the right upper abdomen. The patient had no history of cardiac diseases. Vital signs, urinalysis, hemostasis, prothrombin time, hepatic and renal function, and serum electrolytes were within normal range. He had slight anemia (serum hemoglobin: 11.4 g/dl). White blood cell count and C-reactive protein were elevated to 13600 /μL (normal, 4000–8000 /μL) and 4.0 mg/dl (normal, < 1.0 mg/dl), respectively. His abdominal ultrasound and plain CT demonstrated a large mass (70 mm × 65 mm) between the right hepatic lobe and right kidney (Figure 1). The mass was diagnosed as a ruptured angiomyolipoma at first.

Renal arteriography showed that the inferior adrenal branch of the right renal artery was compressed by tumor; however, pooled contrast fluid and tumor feeding vessels were not identified, and therefore embolization of the artery feeding the tumor was not performed (Figure 2). Contrast-enhanced CT after renal arteriography revealed that the mass had fatty component areas, and that the hematoma existed in peritumoral and perirenal spaces (Figure 3). Abdominal magnetic resonance imaging (MRI) showed a heterogenous mass with fat intensity areas in fat-suppressed T1-weighted images. The peri-tumoral and peri-renal hematoma had higher intensity than the tumor mass and right kidney, suggesting

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case report

Figure 1. Plain abdominal CT shows a large mass (70 mm × 65 mm) in the space between the right hepatic lobe and right kidney at the time of the patient's admission (as shown by the arrow).

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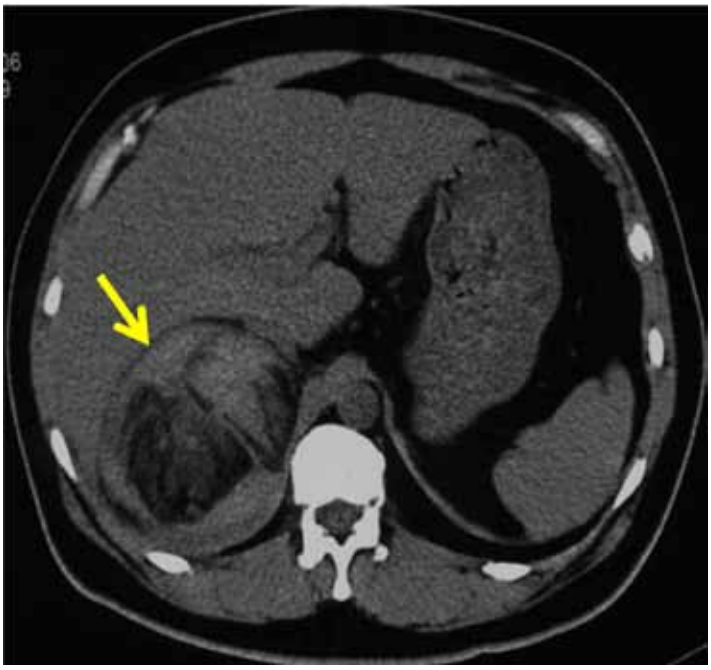

Figure 2. Angiography of the right renal artery. No pooled contrast fluid or tumor feeding vessels were identified. The upper pole of the right kidney was considered to be compressed by tumor (as shown by the arrow).

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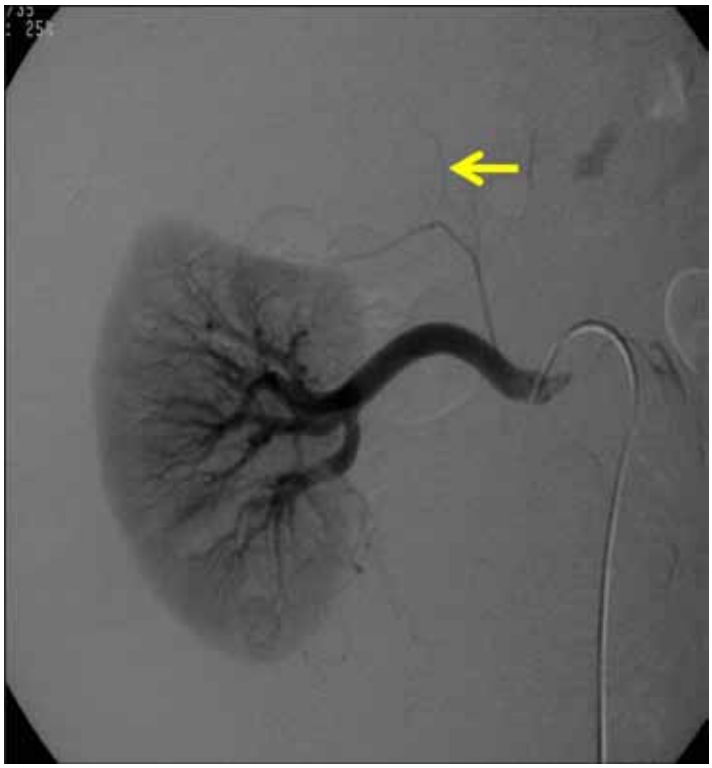

the mass was compatible with right hemorrhagic myelolipoma (Figure 4).

The patient was observed with conservative therapy under hospitalization for 22 days after admission on the watchful waiting plan. Over this period, his symptoms, including flank pain, were diminished. His serum hemoglobin did not drop remarkably. A follow-up CT showed a larger and non-enhanced mass (120 mm × 100 mm × 90 mm) than at the time of admission in the right suprarenal region. The mass had become larger as a result of perinephric hemorrhage (Figure 4). Then radical resection of the right adrenal gland was performed through a midline incision of abdomen. The tumor, hematoma, and adrenal gland were so tightly adherent to the right kidney that their separation was hard to do. The pathological diagnosis was non-malignant adrenal myelolipoma (Figure 5).

DISCUSSION

Adrenal myelolipomas are benign tumors with fat and bone marrow elements. The incidence of this tumor at autopsy is 0.08–0.2% and it consists of mature adipose cells and hematopoietic elements in varying proportions [1, 2]. They are usually asymptomatic, and associated with obesity and hypertension [3]. It is considered a very rare case of retroperitoneal bleeding due to spontaneous rupture of a large adrenal myelolipoma [4] even though adrenal myelolipoma is the most common among benign adrenal soft tissue tumors [5].

Occasionally, they may cause symptoms like flank pain, secondary to compression from tumor bulk, necrosis, and hemorrhage. Small asymptomatic myelolipomas are generally considered candidates for watchful waiting, but symptomatic ones require treatment [6]. They are diagnosed incidentally during imaging by CT or MRI [7]. In general, CT demonstrates fat density with areas of soft tissue. The fatty areas have increased signal intensity, and the areas of bone marrow elements show moderate hyperintensity on T1-weighted MRI [8]. When the mass is detected in the upper pole of the kidney, as we reported,

Figure 3. Abdominal gadrinium-enhanced MRI revealed a heterogenous mass with fat intensity areas on fat-suppressed T1-weighted images. The peri-tumoral and peri-renal hematoma had lower intensity than the tumor mass and right kidney (as shown by the arrow).
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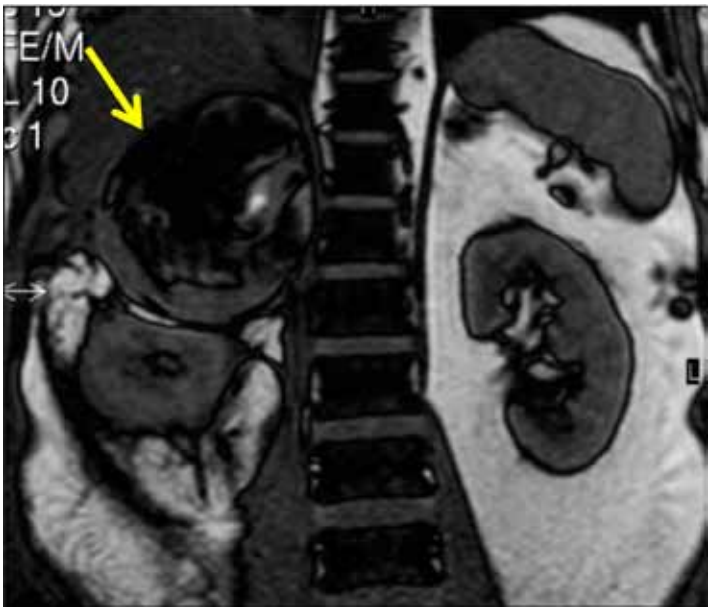

Figure 4. Follow-up abdominal CT showed an enlarged and non-enhanced mass (120 mm × 100 mm × 90 mm) in the right suprarenal region compared with the CT at the time of patient's admission (Figure 1) (as shown by the arrow).
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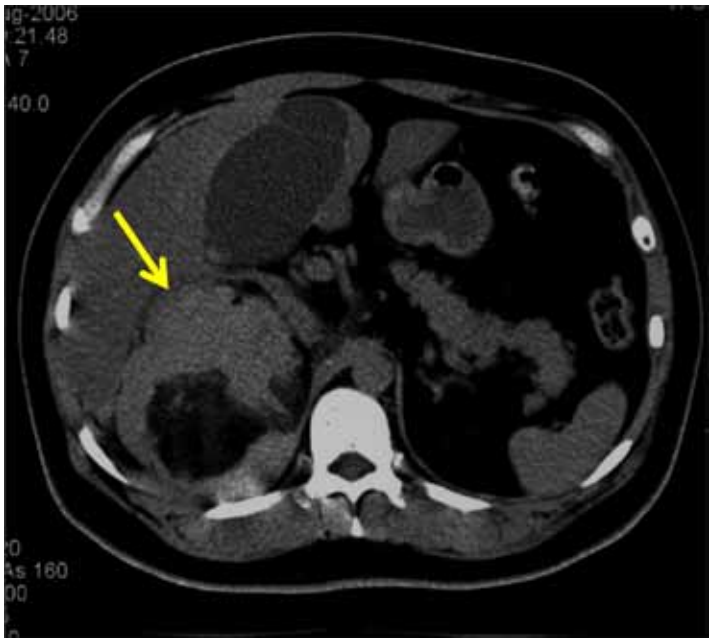

the original organ associated with the tumor can be difficult to define and the differential diagnosis of the retroperitoneal fatty mass such as renal angiomyolipoma, liposarcoma, lipoma, or adrenal myelolipoma [9] is necessary. In our case, CT and MRI demonstrated the fatty areas and the hematoma in the peritumoral and suprarenal space. The margin of the tumor between the liver and the right kidney was sharp and clear on the coronal MRI. Angiography showed a hypovascular tumor. Taken together, we were able to diagnose myelolipoma with retroperitoneal hemorrhage.

This case is the 22nd to our knowledge in which spontaneous rupture of the mass led to retroperitoneal hemorrhage. Characteristics include male dominance (20 of 22) and right-sided predilection (20 of 22), with a mean age of 44 years (range: 20–69) and mean tumor size of 11.7 cm (range: 4–20.5). All known cases were symptomatic tumors. Sudden onset of acute pain is usually due to intratumoral hemorrhage. Extratumoral hemorrhage has been rarely described [10]. In most cases, surgical treatments were performed immediately although spontaneous rupture of the tumor is a rare condition; therefore, the preoperative diagnosis of myelolipoma is important. Imaging including CT and MRI can possibly distinguish hemorrhagic myelolipoma from other conditions such as renal angiomyolipoma, adrenal metastases, pheochromocytoma, and renal adenocarcinoma, which also can cause spontaneous hemorrhage [3, 11, 12, 13]. Arteriography is one of the best methods for accurate diagnosis [14] but can cause severe stress to patients from adverse events. Imaging tests can be simpler and more helpful for the accurate diagnosis of hemorrhagic myelolipoma. In one reported case [15], urgent surgery was needed when the CT showed a myelolipoma hematoma compressing the vena cava. Otherwise, preoperative embolization of ruptured myelolipoma can be useful in stabilizing patients prior to definitive operation [3, 10, 14]. Our case was initially managed by watchful waiting as it was recognized that retroperitoneal hemorrhage was not ongoing. When ruptured myelolipoma is diagnosed, the conservative or watchful waiting approach is recommended because it is safe and less invasive for the patient. When the initial conservative therapy can control the retroperitoneal hemorrhage and maintain a good general condition, an immediate surgical removal with the risk of re-bleeding can be avoided. If an operation is needed due to the failure of conservative treatment of a large adrenal myelolipoma

Figure 5. Histological findings demonstrated that hematopoietic tissue was surrounded with mature fat cells. (hematoxylin & eosin stain. ×200).
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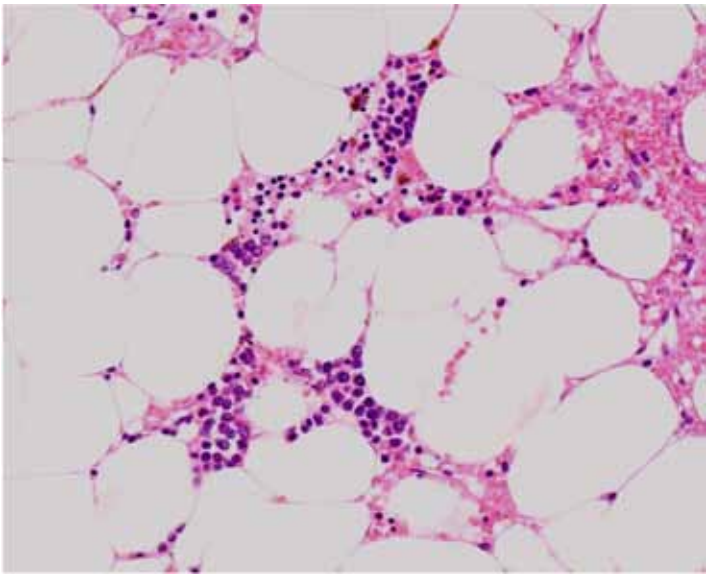

by the watchful waiting approach, an improvement in the patient's general condition is still important before the operation is conducted.

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