

Traumatic Rupture of Renal Angiomyolipoma Managed with Angioembolization Followed by Elective Surgery: A report of Two Cases

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Renal angiomyolipoma (AML) is a rare benign neoplasm of kidney origin. It is mainly comprised of varying amounts of adipose tissue, smooth muscle, and blood vessels. Most patients with small tumors have no clinical symptoms and the tumors are frequently detected by ultrasound or computed tomography (CT).¹ The tumor size is always related to clinical symptoms. Steiner et al. reported that patients with tumor sizes greater than 4 cm were more frequently symptomatic (46%) and required surgery (54%).²

Spontaneous rupture of renal AML is the most severe complication. Koh and George speculated that large intratumoral pseudoaneurysm on contrast-enhanced CT was an important predictor or potential life-threatening tumor hemorrhaging.³ Yamakado et al. demonstrated that groups with spontaneous tumor ruptures have significantly larger sizes of tumors and aneurysm than groups with unruptured ones. Tumor size of 4 cm or larger and aneurysm size of 5 mm or larger were used as predictors of rupture.⁴ In the acute setting of a hemorrhage, transarterial embolization has been used as the treatment of choice for hemostasis in cases of stable hemodynamics.^{5,6} This could provide immediate access to locate the precise site of bleeding and to block the bleeder at the same time.⁶ On the other hand, angioembolization may occlude the aneurysm, destroy solid mass, and eventually, may reduce the size of parts of the tumors.^{2,4,7–8} When the patient's clinical condition was stable after embolization, elective surgery or CT follow-up was recommended.⁹

Regarding the traumatic rupture of renal AML, there have been no reports related to its management. Here, we report two cases of renal AML with traumatic rupture, and suggest the management guidelines for this kind of tumor rupture.

CASE REPORTS

Case 1

A 31-year-old female was struck by a car while she was riding a motorcycle. Two weeks before the accident, she had palpated a mass on her right abdomen and was scheduled to receive radiologic investigation in a hospital. She was initially sent to a local hospital. No specific examinations or treatments were performed there, and she was discharged 6 hours after admission. The next morning she complained of severe pain over her right abdomen and flank. She was immediately brought to our emergency department (ED). On arrival, initial hemodynamic status was stable, but physical examination revealed a huge tender mass palpated on the right side of her abdomen and flank. Gross hematuria was also noted.

Abdominal CT showed a huge mass, which measured 10 × 15 cm and contained adipose tissue, was ruptured with perirenal hematoma and contrast extravasation on the right kidney (Fig. 1). Hypovolemic shock (blood pressure was 73/28 mm Hg) developed 4 hours after her arrival. Resuscitation, crystalloid (1,000 cc) and blood transfusion (red blood cells, 2 units) made her hemodynamics return to normal. Renal angiography was performed immediately. It revealed a space-occupying lesion from the low pole of the right kidney and abnormal hyper-tropic arteries with multiple tiny aneurysms (1–2 mm) and contrast extravasation (Fig. 2). Transcatheter arterial embolization with micro-coil and gel form powder was performed.

Two days after the accident, respiratory distress developed and end tracheal intubation was performed; diffusion haziness of the right lung field was found on chest x-ray film. A chest tube was inserted on the right and 800 cc of blood initially flowed out. Right flank pain was complained of and persistent drop of hemoglobin was noted. Due to suggested ongoing tumor hemorrhaging, repeated angiography (Figure 3) was performed and it demonstrated a small pseudoaneurysm (2 mm) with contrast extravasation of the inferior branch of right renal artery. Micro-coil embolization was performed.

Her condition became stable after two of arterial embolizations. Due to huge and fragile ruptured tumor with indistinct anatomy, elective nephrectomy including the mass was performed 1 month after admission. The pathology demon-

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Fig. 1. A large adipose-containing mass, measured 20 × 15 cm, with perirenal hematoma and contrast extravasation located on the low pole of the right kidney.

strated renal AML. She was discharged 1 week after the operation.

Case 2

A 62-year-old female on foot was struck by a car on her back left side. On arrival to our ED, shock (blood pressure was 86/55 mm Hg) was noted; after resuscitation with crystalloid (Lactate Ringer 2,000 cc) and blood transfusion (whole blood, 2 units), her hemodynamic status returned to normal. Physical examination revealed an ecchymosed on the

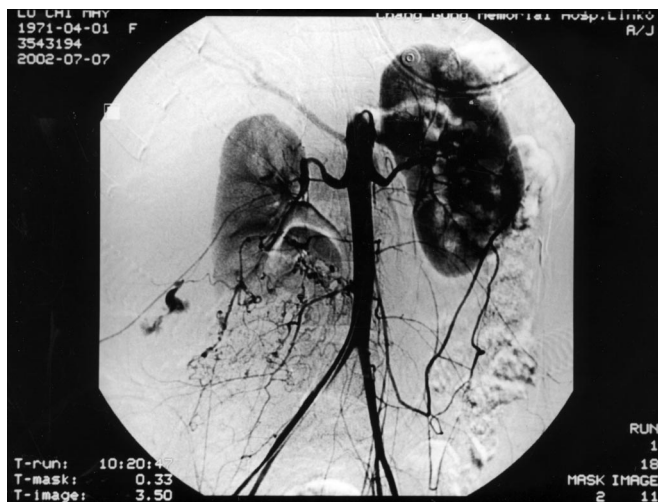


Fig. 2. This shows that a space-occupying lesion with abnormal hypertrophy arteries and multiple tiny aneurysms (1–2 mm) combined with contrast extravasation on the low pole of the right kidney.

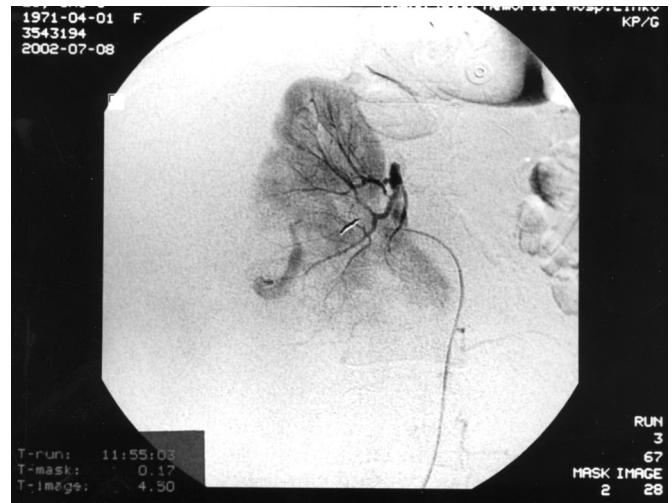


Fig. 3. Repeated angiogram shows that a small aneurysm (2 mm) with contrast extravasation on the inferior branch of the right renal artery.

left side of her back. Abdominal tenderness, especially on the left side of her abdomen, was found.

Emergent abdominal CT (Fig. 4) showed that an adipose-containing tumor (measured 10 × 10 cm) with perirenal hematoma and contrast extravasation was noted at the upper pole of the left kidney. Traumatic rupture of the renal AML was impressed. Emergent angiography of the left renal artery (Fig. 5) showed a huge aneurysm (15 mm) with contrast pooling on the upper pole of the left kidney. Selective embolization with micro coil was performed smoothly. After angioembolization, her condition was stable. She was discharged 2 weeks after angioembolization with restriction of her activities at home. Follow-up CT (Fig. 6) 2 months after



Fig. 4. This shows that an adipose-containing mass with huge perirenal hematoma and contrast pooling, measured 10 × 10 cm, on the upper pole of the left kidney.



Fig. 5. Emergent angiogram shows a huge aneurysm (15 mm) with contrast pooling on the upper pole of the left kidney.

angioembolization demonstrated absorption of the perirenal hematoma, which measured 7.0×7.0 cm. Partial nephrectomy was scheduled. However, attributed to severe adhesion of tumor to surrounding organs and massive hemorrhage from ruptured tumor during manipulation, she eventually underwent left nephrectomy after the image study was performed. She was uneventfully discharged 1 week after surgery. Pathology demonstrated renal AML.

DISCUSSION

Renal AML is composed of varying amounts of three tissues including mature adipose tissue, smooth muscle and



Fig. 6. Follow-up Ct shows that the mass reduced (from 10×10 cm to 7.0×7.0 cm) in size after angioembolization and hematoma absorption at 2 months after angioembolization.

thick walled blood vessels. It predominately occurs in women (74%–94%).^{2,4,10–11} Thirteen of 25% of neoplasm involve bilateral kidneys, in which patients have high rate of tuberous sclerosis.^{2,10} Most patients are asymptomatic, and are often incidentally diagnosed using results of CT, or ultrasound. However, the symptom severity of patients is frequently related to the tumor size. Those with tumor sizes less than 4 cm are less often symptomatic (24%), while those with tumor sizes greater than 4 cm are more often symptomatic (52%).² The most common symptoms of patients with renal AML are abdominal or flank pain (84%), palpable mass (53%), hemorrhage (32%), anemia (21%) and hypertension (21%).²

Spontaneous rupture of renal AML, which is a rare but potentially life-threatening condition, is the most severe complication.^{9–10,12} Ultrasound and CT are the most valuable initial examinations. Ultrasound can easily demonstrate perirenal and subcapsular fluid collections as a screening test. CT can reveal a more definitive and precise image of the exact location and extent of the hemorrhage.^{13–15} In cases of spontaneous tumor rupture with perirenal hematoma, Belville et al. reported that the accuracy of CT for the diagnosis of renal AML was 92%, when a renal mass was demonstrated and interpreted using established criteria.¹⁶ Zagoria et al. recommended that CT should be performed first, then angiogram as the second examination if no mass was detected.¹⁷ If both imaging studies failed to demonstrate a renal mass, then follow-up with thin-slice CT was indicated. Repeat CT may not only allow for resorption of perirenal hematoma and allow for the appearance of a small tumor previously hidden by blood, but also enable differentiation between a benign and malignant neoplasm.^{17–18} Osterling et al. recommended that symptomatic tumors smaller than 4 cm should be observed regularly with CT or ultrasound, while those greater than 4 cm should be studied by angiography and considered for arterial embolization or surgery.¹⁰

The nontraumatic rupture of neoplasm is related to tumor size. As the tumor grows, the blood flow supplying the tumor increases, which leads to vessel dilation and aneurysm formation.^{4,19} On the other hand, rupture of an aneurysm may cause an increase in tumor size. Yamakado et al. reported that aneurysm formation was observed in 22 (76%) of 29 tumors of renal AML, in which 100% of the ruptured tumors had aneurysm formation. The mean tumor size (11.4 cm) and aneurysm size (13.3 mm) were significantly larger than those (5.0 cm and 2.4 mm respectively) in the group with unruptured tumors. Aneurysms formation appeared to be related to tumor size, and large aneurysms had higher probability of rupture.⁴

In cases of acute hemorrhages of tumors, transcatheter arterial embolization has been used as the treatment of choice to stop active bleeding.^{4,14–17,19} The embolization could be useful in preventing growth and spontaneous rupture not only in ruptured tumors but also in unruptured ones.^{4–6,19} Embolization may help prevent tumor growth by resulting in occlusion of the aneurysm, destruction of the solid mass, or

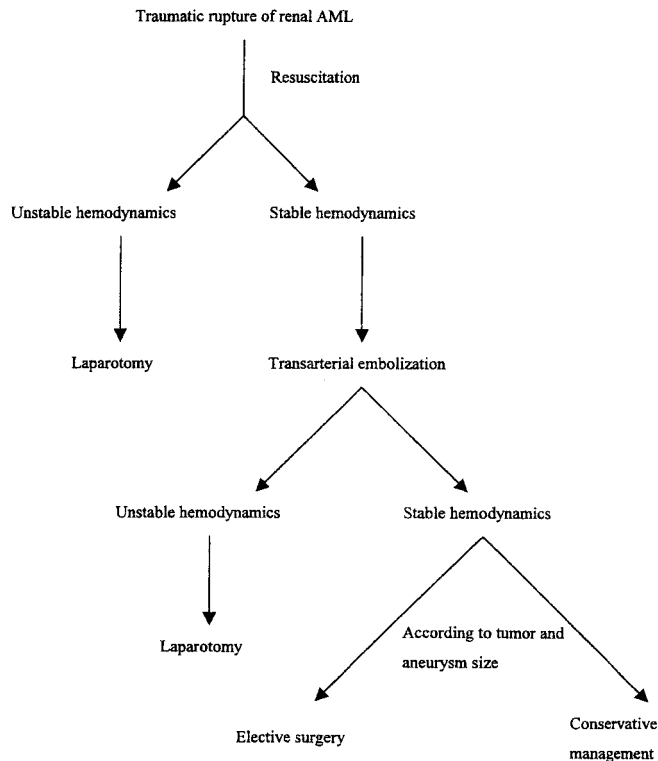


Fig. 7.

both. In spontaneous AML ruptures, most of the cases ranging from 83-100%, were successfully managed with arterial embolization.^{2,4,6,18} The tumor sizes reduced in 57-80% of the tumors and size regression of 13-46% according to long-term follow-up.^{4,6,19}

As far as traumatic rupture of AML, there have been two case reports in the literature,²⁰⁻²¹ but neither concerned the angioembolization management. In our two cases, both had initial hypovolemic shock after trauma. After resuscitation, hemodynamic status soon returned to normal. Initial abdominal CT revealed huge retroperitoneal perirenal hematomas with contrast extravasation. Both were managed with emergent arterial embolization, though the former case needed repeat arterial embolization for aneurysm formation with contrast extravasation. In conditions of emergent laparotomy for ruptured tumor bleeding, there is major blood loss, even uncontrolled hemorrhage during manipulation, in part due to pre-operative massive bleeding, possible ensuing hypothermia, and coagulopathy. This is in part due to the huge fragile tumor and blood clot interfering with the anatomic dissection, and difficulty for hilar accessibility for vascular control. However, transcatheter arterial embolization could localize the bleeding site and perform an embolization for immediate hemostasis during the limited amount of critical time. In cases of stable homodynamic status, the vascular access could provide a more noninvasive and effective method to control acute hemorrhaging of tumors than an emergent laparoscopy could.

After angioembolization, the conditions of both cases stabilized. Owing to the high possibility for rebreeding of the large tumor in the former case and the large aneurysm (15 mm) of the latter case, elective surgery with nephrectomy was performed in both cases. Partial nephrectomy remains the standard of surgical intervention of AML.^{10,22} However, due to enormous fragile ruptured tumor for the former case, and post-embolization severe adhesion of tumor and uncontrolled hemorrhage on manipulation for the latter case, nephrectomies were performed. Both were discharged without complications.

According to our experiences, we designed an algorithm for the management of traumatic rupture of renal AML (Fig. 7). When the hemodynamic status of the patient was persistently unstable after resuscitation, emergent laparoscopy for nephrectomy is mandatory. However, in cases of stable hemodynamic status after resuscitation, emergent transcatheter arterial embolization is indicated for hemostasis. After the angioembolization, when the hemodynamic status of patient is unexpectedly unstable, emergency laparoscopy nephrectomy is mandatory. When the hemodynamic status is stable and there are no signs of further tumor bleeding, elective surgical intervention or conservative management with CT follow-up is chosen according to the tumor size and the aneurysm formation.

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