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Research Letter

Spontaneous rupture of renal angiomyolipoma in the third trimester

Jing Liu [a], Tao Meng [a,*], Xiuhua Yang [a], Ge Zhao [a], Bai Li [b]

[a] Department of Obstetrics and Gynecology, The First Affiliated Hospital of China Medical University, Shenyang, People's Republic of China
[b] Department of Urology, The First Affiliated Hospital of China Medical University, Shenyang, People's Republic of China

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Dear Editor

Angiomyolipoma (AML) is an uncommon benign tumor with the following three histologic features: adipocytes, smooth-muscle cells, and blood vessels with thickened walls [1]. It is more prevalent in women than in men, suggesting that female hormones play a role in its growth. Most renal AMLs are asymptomatic and are found incidentally during imaging examination. Occasionally, however, patients have severe symptoms. The risk of spontaneous rupture during pregnancy is higher than that seen in nonpregnant women, and this complication can be catastrophic [2]. In addition, AML poses a difficult diagnostic and therapeutic challenge during pregnancy. We herein report a rare case of spontaneous renal AML rupture during pregnancy. We would like to remind obstetricians that this neoplasm should be considered when patients present with sudden-onset abdominal and back pain with hypotension or shock.

A 31-year-old primiparous woman presented to our hospital at 37 weeks' gestation with sudden-onset right upper abdominal pain radiating to her back; there were no other symptoms. The entire lower abdomen was tender, but the pain was more severe on the right side. The fetal heart rate was 50–70 beats/min, prompting emergency cesarean delivery for fetal distress. The 1- and 5-minute Apgar scores were 8 and 10, respectively. During surgery, a long, narrow hematoma was noted at the mesentery and in the retroperitoneum. The patient's general condition was good, and her preoperative hemoglobin (Hb) level was 11.0 g/dL, but we opted for a nonexhaustive exploration of the retroperitoneal space due to the risk of rupture associated with handling this hematoma. The cesarean was completed, and computed tomography (CT) was suggested to the patient, but she declined. Her postoperative clinical status was stable, and we continued to monitor her physical signs and Hb level closely: there was no change in her postoperative Hb level (10.5 g/dL) over the first 36 hours. Two days later, her clinical status remained stable, but her Hb level dropped to 7.1 g/dL. At this point, we persuaded her to undergo immediate abdominal CT, which revealed a suspected AML at the right kidney. A hyperdense area surrounded the kidney, suggestive of hematoma (Figure 1). The patient subsequently became hemodynamically unstable and was taken to the operating suite for immediate nephrectomy. The urologist explored the retroperitoneal area and confirmed an 8 cm × 6 cm renal mass with rupture of the renal capsule. Pathologic examination of the specimen confirmed the diagnosis of AML. The patient had an uneventful recovery from this second surgery.

Renal AML is usually asymptomatic, but it can manifest as flank pain, a palpable abdominal mass, hematuria, or nausea and vomiting. The risk of rupture increases with tumor size, the presence of symptoms, the presence of tuberous sclerosis, and pregnancy. The precise reason that AML is particularly susceptible to rupture during pregnancy has not been confirmed, but might be related to the hemodynamic changes of pregnancy. Blood vessels may weaken when subjected to the burden of increased cardiac output, increased renal blood flow, elevated systemic blood pressure, and increased steroid production [3]. The presence of acute abdominal or lumbar pain, hypotension, or shock should suggest the possibility of tumor rupture. Obstetricians unfamiliar with the condition are likely to make an inappropriate diagnosis such as amniotic-fluid embolism, pre-eclampsia, placental abruption, or pulmonary embolism [4]. We initially diagnosed a placental abruption in our patient, even though she did not have any risk factors for this complication of pregnancy.

Ultrasonography of the abdomen and pelvis may detect a hematoma, but this imaging modality is not precise enough to ascertain the cause. CT is the gold standard, as it is highly sensitive and is able to determine the type, site, and extent of hematoma. It can also detect the presence of fat in the tumor, which is highly suggestive of AML [5]. Magnetic resonance imaging can also differentiate fat by its high signal intensity and is a complementary examination; it is used when CT is contraindicated and when lesions are small or complex.

The management of AML has been widely discussed in the literature. Asymptomatic tumors smaller than 4 cm should be followed with periodic ultrasounds and CT examination every 6

* Corresponding author. Department of Obstetrics, The First Affiliated Hospital of China Medical University, Shenyang 110001, People's Republic of China.
E-mail address: mengtao201011@163.com (T. Meng).

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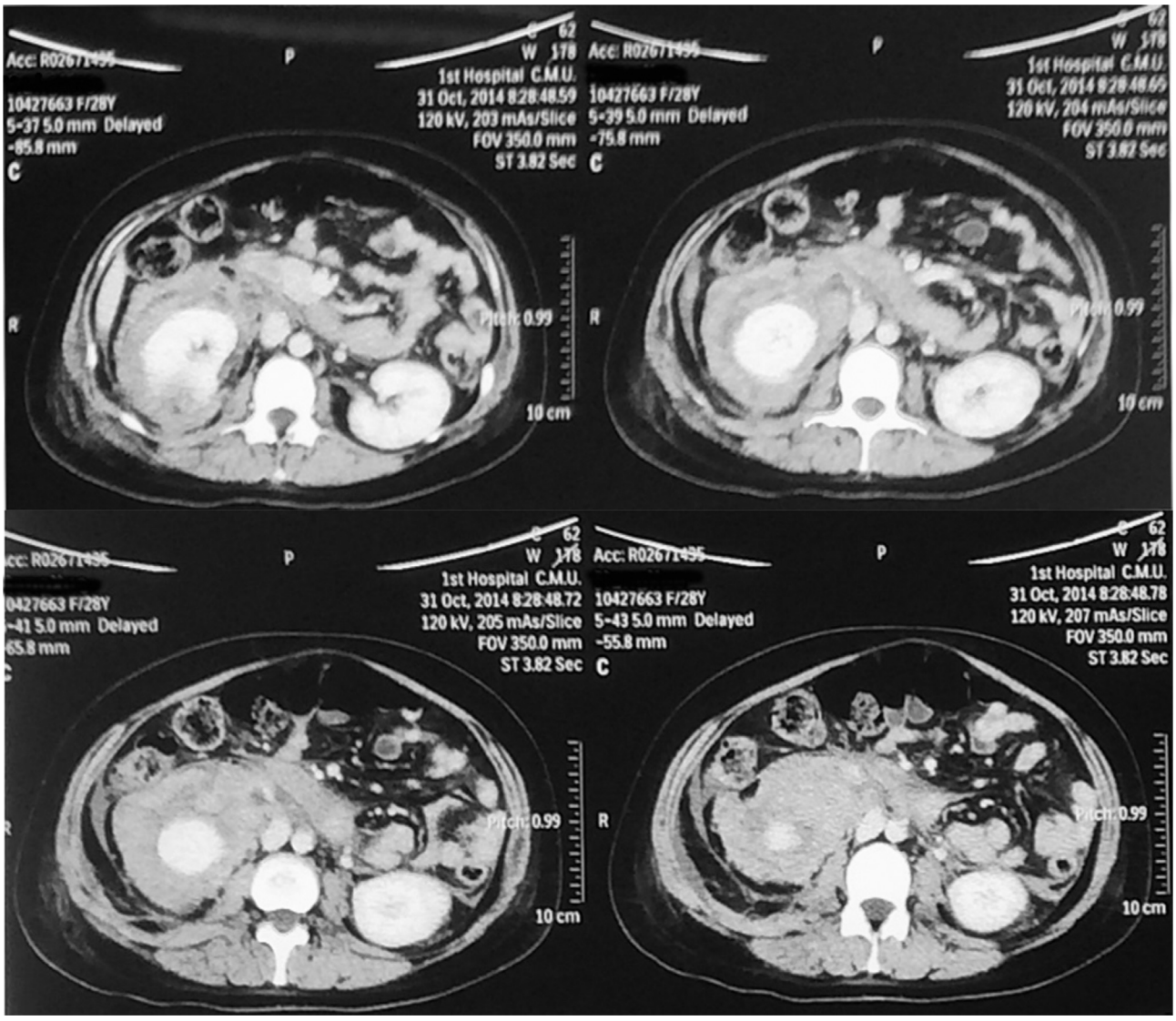

Figure 1. Computed tomography image showing a hematoma around the right kidney.

months [6]. Symptomatic, bilateral lesions should be treated with selective arterial embolization or partial nephrectomy. Radical nephrectomy is required when the patient is hemodynamically unstable. Embolization is the treatment of choice in most cases, as this usually allows for greater preservation of renal function, and is of particular importance for young women. However, recurrence is common, and follow-up surveillance is required [7]. Although embolization of AML during pregnancy has been reported, it is difficult to quantify the long-term effect of radiation on the fetus [8].

When renal AML is diagnosed, the mode and timing of delivery should be individualized to achieve the best outcome for both mother and fetus. The obstetrician must take into consideration the gestational age, the patient's symptoms, the size of the tumor, and the presence or absence of bleeding within the neoplasm. There is a single reported case of a patient at 18 weeks' gestation with low back pain radiating to the right upper abdominal quadrant. Ultrasound suggested a hyperechoic, heterogeneous AML, and probable hemorrhage. The patient underwent conservative management, and the bleeding resolved. Because she was hemodynamically stable and the bleeding stopped spontaneously, she was expectantly managed. At 35 weeks' gestation, she underwent cesarean section for frequent and progressively worsening pain [9]. A planned delivery, either by cesarean or induction of labor, will ensure that multidisciplinary staff is available and prepared to deal promptly with any complications. Although it seems intuitive that the variations in blood pressure experienced during labor and the Valsalva maneuver required for pushing could increase the risk of potentially fatal bleeding, there is no evidence that a higher incidence of bleeding occurs during vaginal delivery than with cesarean.

Urologists should inform female AML patients that pregnancy can increase the risk of rupture, and they should be certain that patients are familiar with the associated symptoms. Patients with known AMLs need to inform their obstetrician when establishing antenatal care. These precautions will help obstetricians to make appropriate and timely diagnosis and management decisions. In conclusion, we remind obstetricians to consider the possibility of this rare disease when patients have sudden-onset abdominal pain and hypovolemic shock. After timely and precise diagnosis, individual management decisions should be made for each patient.

Conflicts of interest

The authors have no conflicts of interest to declare.

References

[1] Kontos S, Politis V, Fokitis I, Lefakis G, Koritsiadis G, Simaioforidis V, et al. Rapture of renal angiomyolipoma during pregnancy: a case report. Cases J 2008;1:245.
[2] Lim CH, Mulvin D. Embolisation of bleeding renal angiomyolipoma in pregnancy. Open J Urol 2011;1:25–7.
[3] Soliman KB, Shawky Y, Abbas MM, Ammary M, Shaaban A. Ruptured renal artery aneurysm during pregnancy, a clinical dilemma. BMC Urol 2006;6:22.
[4] Rafi J, Muppala H. Retroperitoneal haematomas in obstetrics: literature review. Arch Gynecol Obstet 2010;281:435–41.
[5] Cifuentes M, Calleja F, Hola J, Daviú A, Jara D, Vallejos H. Renal angiomyolipoma rupture as a cause of lumbar pain: report of one case. Rev Med Chil 2008;136:1031–3 [in Spanish].
[6] Dickinson M, Ruckle H, Beaghler M, Hadley HR. Renal angiomyolipoma: optimal treatment based on size and symptoms. Clin Nephrol 1998;49:281–6.
[7] Kothary N, Soulen MC, Clark TW, Wein AJ, Shlansky-Goldberg RD, Crino PB, et al. Renal angiomyolipoma: long-term results after arterial embolization. J Vasc Interv Radiol 2005;16:45–50.
[8] Morales JP, Georganas M, Khan MS, Dasgupta P, Reidy JF. Embolization of a bleeding renal angiomyolipoma in pregnancy: case report and review. Cardiovasc Intervent Radiol 2005;228:265–8.
[9] dos Santos MM, Proença SM, Reis MI, Viana RM, Martins LM, Colaço JM, et al. Spontaneous rupture of renal angiomyolipoma during pregnancy. Rev Bras Ginecol Obstet 2014;36:377–80.