

Urgent Angioembolization With Early Elective Nephron-sparing Surgery for Spontaneously Ruptured Renal Angiomyolipoma

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Tumor rupture with hemorrhage is a severe complication of renal angiomyolipoma. Urgent transcatheter arterial embolization is an effective method for the patient suffering from life-threatening hemorrhage due to ruptured angiomyolipoma. Elective surgery has been suggested because of the possibility of rebleeding or abscess formation after embolization. However, it is not easy to perform a nephron-sparing procedure by delayed elective surgery due to severe tissue adhesion after bleeding. Herein, we report 2 cases of ruptured renal angiomyolipoma initially managed with transcatheter arterial embolization. Nephron-sparing surgery was performed a few days after embolization, before tissue adhesion occurred. Both kidneys were preserved, and the postoperative course was uneventful. [*J Chin Med Assoc* 2009;72(8): 450–452]

Key Words: angiomyolipoma, embolization, nephron-sparing surgery

Introduction

Renal angiomyolipoma (AML) is a benign neoplasm containing various amounts of adipose tissue, smooth muscle, and vascular tissue. AML accounts for 0.3–3% of renal tumors and 1% of surgically resected neoplasms.¹ Most AMLs are asymptomatic and are diagnosed incidentally. The most severe symptom is related to rupture of AML, which may lead to life-threatening hemorrhage. Most patients with life-threatening hemorrhagic AML require total nephrectomy if it is explored. With the advent of transcatheter arterial embolization (TAE), the technique has been used to treat acute hemorrhage caused by ruptured AML. TAE may stop renal hemorrhage effectively and safely by precise localization of the bleeding points. However, after embolization, tumors shrink with liquefactive necrosis and tend to induce abscess formation or other complications. In order to prevent these complications, delayed elective surgery after hemodynamic status has been stabilized by TAE has been suggested by some authors.² However; such surgery faces the problem of severe tissue adhesion, which may lead to inevitable nephrectomy.

In this report, we present our experience with early elective nephron-sparing surgery a few days after TAE in 2 patients. Both patients tolerated the surgery very well, and the affected kidneys were preserved without any sequelae.

Case Reports

Case 1

A 46-year-old woman was referred to our emergency department due to severe left abdominal pain for 1 day. On arrival, she was normotensive with a rapid pulse (124/min). Physical examination revealed a huge, tender palpable mass over the left upper abdomen. Her hemoglobin level was 9.1 g/dL. Abdominal computed tomography (CT) demonstrated a large heterogeneous fat-containing tumor over the upper pole of the left kidney with diffuse infiltrative high-density lesion in the perirenal space (Figure 1). A large AML with bleeding was considered. Emergent renal angiography revealed abnormal tumor vessels with multiple aneurysm formation (Figure 2). A catheter was put



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E-mail: b729@cych.org.tw • Received: November 13, 2008 • Accepted: May 6, 2009

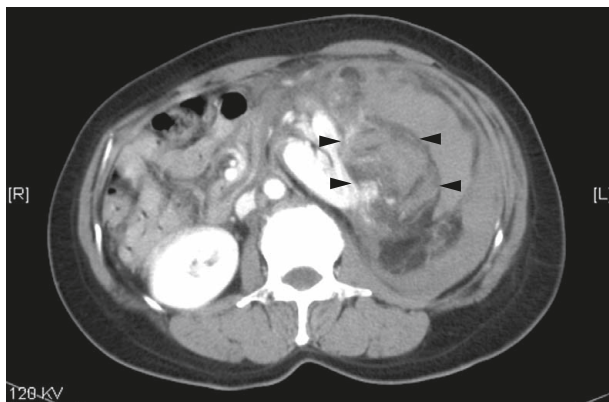


Figure 1. Abdominal computed tomography demonstrates a large heterogeneous tumor (arrowheads) with fat content over the upper pole of the left kidney, which is diffusely infiltrated by hemorrhage.



Figure 2. Renal angiography reveals abnormal tumor vessels (arrowheads) with tumor stain and multiple aneurysm formation in the tumor mass.

in the feeding artery, and absorbable gelatin sponge pieces were injected to block the feeding blood flow. The condition became stable after embolization. Low grade fever (up to 38°C) was noted for 3 days after embolization. The abdominal pain subsided gradually after embolization, and there was no other post-embolic symptom.

Elective surgical exploration was performed 1 week after embolization. Sloughed tumor fragments mixed with blood clots within Gerota's fascia were noted during exploration. The tumor was enucleated, and the blood clots removed. The tumor fossa was sutured with catgut. Blood loss was minimal, and renal pedicle control was not necessary because the feeding artery had already been obliterated by TAE. During the course of the whole operation, the surgical plane was clear without severe adhesion.

The pathologic finding confirmed the diagnosis of AML. After operation, the patient recovered well and was discharged 1 week later. She was followed-up regularly with renal ultrasound; no rebleeding, tumor recurrence or kidney atrophy was noted postoperatively.

Case 2

A 49-year-old woman visited our emergency department due to right groin and lower abdominal pain for 2 days. Her vital signs were stable. She had pale conjunctiva, and her hemoglobin level was 6.3 g/dL. Physical examination revealed right flank knocking pain and tenderness over the right lower abdomen. Abdominal CT showed a 10-cm mass lesion over the lower pole of the right kidney with fat density and a focal area of hyperdensity. Hemorrhagic AML was impressed. Urgent renal angiography revealed a large hypervascular stained tumor with bleeding. The feeding artery was embolized with absorbable gelatin sponge pieces. The post-embolic course was smooth except for mild fever that lasted for 2 days.

The patient then underwent elective nephron-sparing surgery 7 days after TAE, with operative procedures and findings similar to those in Case 1. The postoperative course was uneventful and postoperative ultrasound demonstrated normal-sized kidney with no rebleeding or tumor recurrence.

Discussion

The clinical signs of AMLs are related to the tumor size, and most patients with small AMLs have no clinical symptoms. If AML size is ≥ 4 cm, 80–90% of the tumors will be symptomatic, and 50–60% of them will bleed spontaneously.¹ Spontaneous rupture is the most severe complication of AMLs that are ≥ 4 cm in size. The optimal treatment of choice for AML is related to its size. Dickinson and colleagues reviewed 48 renal units and demonstrated that small asymptomatic lesions (< 4 cm) tend to remain stable but should be periodically evaluated. Medium-sized lesions (4–8 cm) have the most variable behavior. These lesions should be followed closely with serial imaging studies, and if significant changes in size or symptoms are noted, or the patient is at risk for flank trauma, elective intervention should be initiated promptly to increase the chances of renal salvage. Large asymptomatic angiomyolipomas (> 8 cm) will most likely become symptomatic and should be treated electively prior to the development of symptoms and potential complications.³

In the past, the standard treatment for AML was radical nephrectomy, because of the inability to make

a precise preoperative diagnosis and the possibility of massive hemorrhage.⁴ With improvements in surgical skills and technology, nephron-sparing procedures, such as partial nephrectomy, tumor enucleation and angioembolization have emerged as alternative methods of AML treatment. In 1977, Moorhead et al reported the first successful experience of using superselective percutaneous TAE to manage renal hemorrhage secondary to AML in a solitary kidney.⁵ Since then, serial studies about the safety and efficacy of superselective TAE treatment for ruptured AML have been reported. With its minimal potential for damage to normal renal parenchyma, superselective angioembolization is now the treatment of choice for life-threatening renal hemorrhage. A variety of embolic agents, including coils, liquid agents, and polyvinyl particles, have been used for angiographic renal tumor embolization. However, there is no clear consensus regarding the embolic agent of choice.⁶ The complication rate of TAE is 10%, with the most common complication being abscess formation (5%). Recurrent hemorrhage, re-embolization, and necessity for further surgical intervention were noted in 17%, 14% and 16% of patients, respectively. Further surgical interventions are performed usually for recurrent bleeding or persistent symptoms.⁷ The pitfall is tissue adhesion after renal hemorrhage, which makes nephron-sparing surgery more difficult and sometimes leads to inevitable renal loss.^{2,8} Both of our patients underwent early nephron-sparing surgery after TAE with good results. Due to limited case experience, the proper timing for surgical intervention after tumor embolization is uncertain. Adhesions will form within 72 hours after tissue injury and their formation is more intensive at about 10 days to 2 weeks.⁹ It seems reasonable to perform nephron-sparing surgery within 1 week after tumor embolization. The advantages of early post-TAE surgical intervention include less surgical and anaesthetic

risk with stabilized hemodynamic status, decreased renal bleeding and no need for renal pedicle control during operation, avoiding the possible complications of retained blood clots and necrotic tumors in the retroperitoneal space, less tissue adhesion than that confronting delayed elective surgery, and obtaining pathologic diagnosis. However, due to limited case number in the present report, collection of more patient data is necessary to confirm the safety and efficacy of this modality of treatment for severe hemorrhage due to ruptured AML.

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