

Case Report

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Urgent arterial embolization of ruptured renal angiomyolipoma

Abstract: The most undesired complication of renal angiomyolipoma (AML) is bleeding. Because of tumor rupture, the bleeding can spread to the retroperitoneal field and can be severe enough to be life threatening. We report a case of retroperitoneal hemorrhage caused by a ruptured AML that was successfully treated with transarterial embolization with N-butyl cyanoacrylate.

Keywords: Hemorrhage; kidney; therapeutics; neoplasms; angiomyolipoma; embucrilate

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1 Introduction

Renal angiomyolipoma (R-AML) is a benign tumor consisting of different ratios of blood vessels, adipose tissue, and smooth muscle tissue. These tumors are not rare, they are seen twice as much in females, and they are mostly small and asymptomatic when detected [1]. R-AML is reported to grow faster during pregnancy, which is related to increasing hormonal influence [2,3]. R-AML is usually identified when the patients are being examined for other complaints. The clinical importance of R-AML is related to bleeding caused by the abnormal vascular structures it contains [4]. The arteries contained in R-AML have extraordinary configurations and are numerous and tortuous. Histologically, these vessels do not have internal elastic lamina and their smooth muscle layer is disrupted, which is why they are prone to aneurysm formation and rupture [4,5]. When the tumor size exceeds 4 cm, most of the

patients develop symptoms related to tumor bleeding and mass effect [5]. In the treatment of R-AML, if the lesions greater than 4 cm are asymptomatic the recommendation is to have radiological follow up every six months. If they are symptomatic, endovascular or surgical therapy is recommended after this [6]. For asymptomatic lesions that are greater than 8 cm, the possibility of developing symptoms is high, and treatment should be performed [7]. The most undesired complication of R-AML is bleeding. When the bleeding is intra-tumoral, it can either be limited or can spread to the intra-peritoneal field as a result of tumor rupture and be severe enough to threaten life. We report a case of retroperitoneal hemorrhage caused by a ruptured R-AML that was treated with transarterial embolization with a non-reported embolic material for R-AML.

2 Case report

A 38-year-old male patient was admitted with the complaints of left flank pain and hematuria. The patient did not have any history of trauma; however, in his physical examination, there was tenderness on palpation on the left side. The laboratory findings were within normal limits: hemoglobin was 13.4 g/dl and hematocrit was 40.2%. Abdominal computed tomography (CT) examination performed with intravenous (IV) contrast material injection identified a heterogeneous mass of 9.5 x 6 cm on the left kidney that contained adipose tissue and a large field of hematoma that extended to the retroperitoneal field (Figure 1). Through CT findings, a diagnosis of retroperitoneal bleeding due to an R-AML rupture was established. During 18 hours of follow up, the patient developed tachycardia and hypotension; hemoglobin dropped to 9.9 g/dl and hematocrit to 29.9%. The patient described severe pain, irritation, and sweating. As he had a decreasing ability to cooperate and increasing instability, a decision was made to perform an emergency transarterial embolization treatment. Ceftriaxone 1 gr and gentamycin

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Figure 1: Abdominal CT demonstrates a large retroperitoneal hematoma due to left R-AML rupture

160 mg were administered for prophylactic purposes and the procedure was initiated.

Digital subtraction angiography was performed using a right common femoral artery approach. First, diagnostic abdominal aortography was performed to localize the renal arteries and to investigate the feeding arteries other than the renal artery. There were two left renal arteries originating from the abdominal aorta. Both left renal arteries were selectively and individually catheterized with 5F diagnostic renal catheter, and the images obtained showed that the mid-zone segmental artery that originated from the renal artery on the top was feeding the R-AML; this artery was identified as having a pseudoaneurysm and extravasation (Figures 2 and 3). The segmental artery feeding the R-AML was catheterized superselectively with the microcatheter passed through the renal catheter. Because of the extravasation of the contrast material, which meant active hemorrhage, the worsening of the patient's general condition, and his hemodynamic instability, we preferred to use N-butyl cyanoacrylate (NBCA) glue for embolization for rapid recovery. The lumen of the microcatheter was washed with 5% glucose solution and a mixture consisting of 1 mL NBCA glue (histoacryl, Braun), and 2 mL iodized oil (lipiodol ultra-fluid, Guerbet) was carefully injected into the feeding artery of the R-AML under fluoroscopic control (Figure 4). When an occlusion was seen in the feeding artery, the microcatheter was rapidly retrieved and removed from the renal catheter. To ensure that there is no glue within the lumen of the renal catheter, blood was aspirated. In the control angiography following embolization, we confirmed that the feeding artery of the R-AML



Figure 2: Selective angiogram of left upper renal artery demonstrates pseudoaneurysm in the tumor



Figure 3: Late arterial phase of left upper renal artery demonstrates abnormal vascularity of AML, and extravasation from the pseudoaneurysm

was occluded completely and that the pseudoaneurysm and the extravasation had disappeared (Figure 5). It was also seen that a small branch other than the segmental branch that was feeding the mass was also occluded. Following endovascular treatment, the general condition of the patient improved rapidly. The patient refused the complementary nephron-sparing surgery and he was discharged after five days. The patient did not have any complaints during his three month follow up period. He came to control two years later. The contrast CT examination performed 31 months after the procedure showed that the

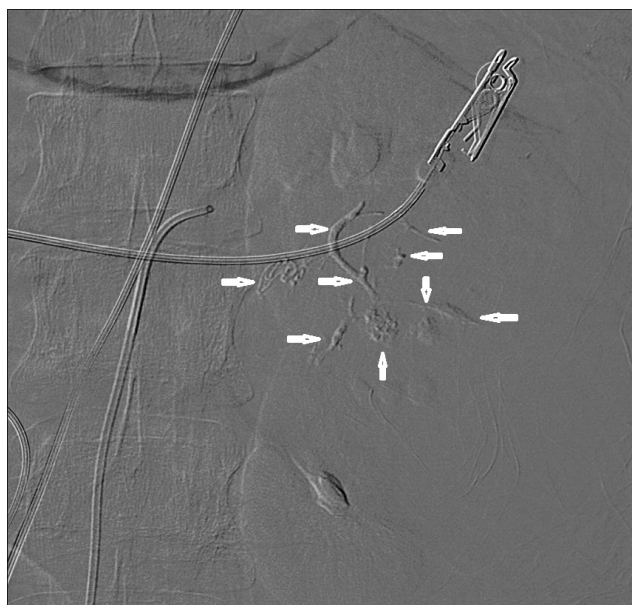


Figure 4: Angiogram before the contrast material injection shows glue cast (arrows)



Figure 5: Control angiogram after the embolization, demonstrates occlusion of the feeding artery of the AML with disappearance of the pseudoaneurysm and the tumor vascularity. It also demonstrates occlusion of a small branch other than the feeder of the AML (arrow)

size of R-AML regressed to 4.4 x 3.5 cm, and there was no significant contrast enhancement inside the mass related to vascular structures or solid components (Figure 6).

Ethical approval: The research related to human use has been complied with all the relevant national regulations, institutional policies and in accordance the tenets of the Helsinki Declaration, and has been approved by the authors' institutional review board or equivalent committee.



Figure 6: CT examination performed 31 months after the embolization reveals regression of the R-AML size

Informed consent: Informed consent has been obtained from all individuals included in this study.

3 Discussion

The most severe complication of R-AML is bleeding, which can be life threatening. The bleeding in R-AML can be spontaneous or can follow a trauma. Nearly half of the cases that are larger than 4 cm develop spontaneous bleeding [5]. The main factors that increase the risk of bleeding are the size of the tumor at the time of diagnosis, size increase in a tumor that is under follow up, and a symptomatic tumor and tuberous sclerosis accompanying R-AML [8–11]. For this reason, tumors larger than 4 cm should be followed up at shorter intervals than those smaller than 4 cm. Follow up for symptomatic tumors smaller than 4 cm should occur more often than asymptomatic tumors smaller than 4 cm. Follow up should happen in cases with tuberous sclerosis more often than those who do not have tuberous sclerosis. Asymptomatic tumors larger than 8 cm, symptomatic tumors larger than 4 cm, asymptomatic tumors that are larger than 4 cm and that also have tuberous sclerosis and the tumors that show an increase in the size during follow up should be treated with endovascular therapy or surgery under elective conditions [6–8,10].

Radiological modalities used in the evaluation of R-AML include ultrasound, CT, and magnetic resonance imaging. Ultrasound alone cannot be used in the diagnosis of R-AML because renal cell carcinoma can also be hyperechoic just as R-AML [12]. R-AML is usually diagnosed

using CT and magnetic resonance imaging. Color Doppler ultrasound can assist in the diagnosis of pseudoaneurysm in the tumor and occlusion of the pseudoaneurysm after embolization. Color Doppler ultrasound can also be used for the assessment of renal arterial hemodynamic changes [13].

For bleeding R-AML, main treatment alternatives are conservative, transarterial embolization, partial or total nephrectomy, and immediate or delayed surgery after embolization. Because of the unfavorable effects of angiography and embolization and surgical treatment on a fetus, hemodynamically stable pregnant women should be treated with conservative treatment and the tumor should be treated after delivery [2]. For bleeding R-AML, urgent total nephrectomy can be performed; however, in hemodynamically unstable patients, risk of complication increases and healthy renal tissue can be unnecessarily excised together with the tumor tissue. In patients with impaired or limited renal functions, it is very important to preserve healthy renal tissue. If the tumor has not totally invaded the kidney, total nephrectomy should not be considered. In unstable patients, in the presence of a large size hematoma, nephron-sparing surgery is considerably difficult [14]. For this reason, despite being treatment alternatives for bleeding R-AML patients, total and partial nephrectomy should be performed under elective conditions. Surgical treatment can be more appropriate after stopping the bleeding with endovascular treatment and rendering the patient more stable. The recommendations are such that nephron-sparing surgery should be performed within the week following the embolization [15]. Transarterial embolization is a preferred treatment alternative for renal bleeding and is used in a widespread manner for R-AML treatment. Embolization is a normal kidney tissue preserving, minimally invasive treatment alternative that provides us with an expedited intervention opportunity for bleeding and unstable R-AML patients.

It is important to know the limitations and complications of transarterial embolization. The main limitations are contrast material allergy, kidney function impairments, coagulation disorders, the presence of infection, and vascular anatomy not amenable to superselective tumor embolization. The complications of endovascular treatment can develop due to femoral approach, catheterization, and embolization. The main complications are bleeding, dissection, occlusion, outside target embolization, and post-embolization syndrome. Furthermore, there might be necrosis of the tumor, and, in the event of infection, abscesses might develop. If abscesses develop, percutaneous drainage or nephrectomy might be required.

For transarterial embolization, several embolizing agents like absolute alcohol, gelatin sponges, polyvinyl alcohol (PVA) particles, embospheres, coil, absolute alcohol and iodized oil, ethanol and PVA, PVA and coil, PVA and gelatin sponges, and an amplatzer vascular plug have been used separately or in combination [4,5,8,14,15]. Absolute alcohol, ethanol, gelatin sponges, and PVA and embosphere are agents for distal embolization. During some of the transarterial embolization cases performed with alcohol and PVA, the aneurysm inside the tumor was reported to rupture. This was thought to be related to the increase of pressure inside the aneurysm when the flow inside the feeding artery was being guided towards the aneurysm following the distal embolization of the tumor tissue [4,16]. In embolizations performed with gelatin sponges, there is a high possibility of recanalization. A coil and amplatzer vascular plug is an agent for proximal occlusion. Because of the previously mentioned disadvantages, in a study of the endovascular treatment of R-AML, proximal embolization was conducted with coil after the distal embolization with PVA [4]. Using the same logic, combining several distal and proximal occlusion materials can result in more effective results in the treatment of R-AML.

NBCA glue is an embolizing agent used in vascular embolizations; however, its use has not been reported for R-AML embolizations. In a hemodynamically unstable patient of ours, whose general condition deteriorated in the angiography unit, after diagnosing bleeding during diagnostic angiography, we performed embolization with NBCA glue to obtain a rapid treatment response. NBCA glue is prepared by mixing the glue with iodized oil. As the iodized oil/glue ratio increases, the polymerization time of the glue lengthens, and its viscosity increases. In our case, we wanted to have low viscosity to prevent the escape of the NBCA glue to the outside of the tissue, so an iodized oil /glue was prepared at a 2:1 ratio. The feeding artery and its branches inside the tumor and the pseudoaneurysm were embolized. Thereby, a large vascular system was embolized and a proximal dominant tumor embolization was performed. We did not choose to use alcohol, PVA, or embospheres as these materials could extravasate rather than reach the tumor because of bleeding, and there would be a possibility of severe complications that prohibited effective treatment. The reason for not preferring coil embolization was that it would make possible a shorter segment occlusion, provide a treatment response that would be obtained over a longer period, and run a higher risk of recanalization. Moreover, the cost of the treatment to be performed with coil would be more expensive than NBCA glue. The insight this case provided

us with is that in R-AML without active bleeding, distal and proximal embolization should be performed in the short term by containing the aneurysm and using high iodized/glue ratio to increase NBCA glue viscosity.

Most important complications of NBCA lie outside the target embolization due to the reflux of the embolic agent and the sticking of the catheter tip to the glue cast inside the vessel. Although there was no glue reflux during the embolization, there was occlusion of a small renal artery branch outside the target renal artery branch that was feeding the tumor in the control angiography. During the retrieval of the microcatheter, a small, undetected piece of glue that was stuck to the tip of the catheter going to that branch was thought to be a possible cause of this occlusion.

Patient follow up after embolization can be conducted using laboratory findings, ultrasonography, and CT or magnetic resonance images. The laboratory findings of our patient were stable. In the control CT examination performed 31 months after the procedure, there was only adipose tissue inside the shrinking tumor; the size of the tumor was not decreasing, and this was linked to adipose tissue dominance. In IV contrast CT images, no contrast enhanced vascular or soft tissue density components were identified, and, as the patient was asymptomatic, additional endovascular or surgical treatment was not considered.

4 Conclusion

A R-AML rupture might result in severe enough bleeding to necessitate emergency intervention. Transarterial embolization for a R-AML rupture is an effective, safe, and minimally invasive treatment technique that can be performed by using NBCA glue as the embolizing agent.

Conflict of interest: The authors declare that they have no conflicts of interests.

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