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Case Report

Embolization of hemorrhagic renal angiomyolipoma complicated by arteriovenous shunting: A case report

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ABSTRACT

Arteriovenous shunting associated with angiomyolipoma is an unusual entity, which carries important implications to embolization approach. We present a distinctive case involving a 41-year-old woman who presented with retroperitoneal hemorrhage relating to renal angiomyolipoma. During angiography for urgent embolization, a complex vascular supply with arteriovenous shunting was encountered. Superselective embolization using alcohol or small particles is the standard approach to definitive treatment of symptomatic angiomyolipoma; however; their use is precluded in the setting of arteriovenous shunt hemodynamics. In this case, a 2-step approach was employed by which the initial hemorrhage was treated with proximal embolization using large gelatin foam and metallic coils. This resulted in decreased flow through the arteriovenous shunt, allowing the use high viscosity ethylene vinyl alcohol copolymer dissolved in dimethyl sulfoxide for definitive treatment.

Case review

The patient is a 41-year-old woman with a history of renal angiomyolipomas (AML) who presented to the Emergency

Compliance with Ethical Standards:
(1) This study was not supported by any funding.
(2) The authors declare that they have no conflict of interest.
(3) For this type of study, formal consent is not required.
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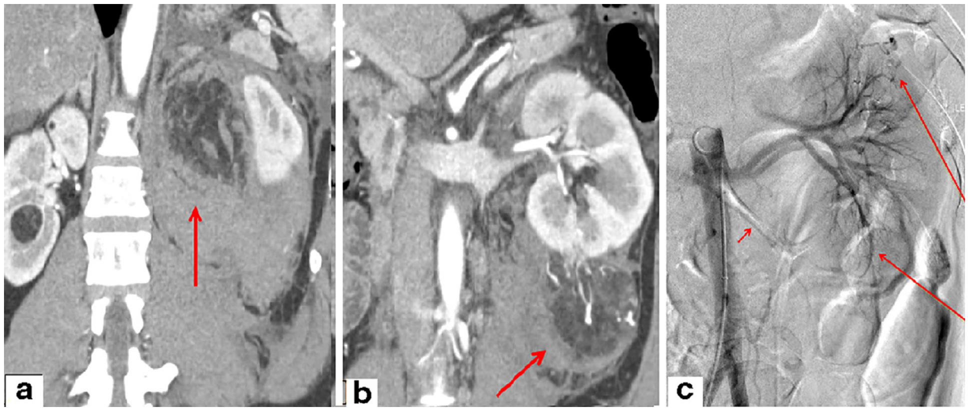

Fig. 1 – (a,b) Coronal CT images showing 2 large AMLs in the upper and lower left renal poles, with retroperitoneal hemorrhage. (c) Abdominal aortography demonstrating left renal artery branches (long red arrows), and a small vessel arising from the aorta, directed toward lower pole lesion (short red arrow).
AML, angiomyolipomas; CT, computed tomography.

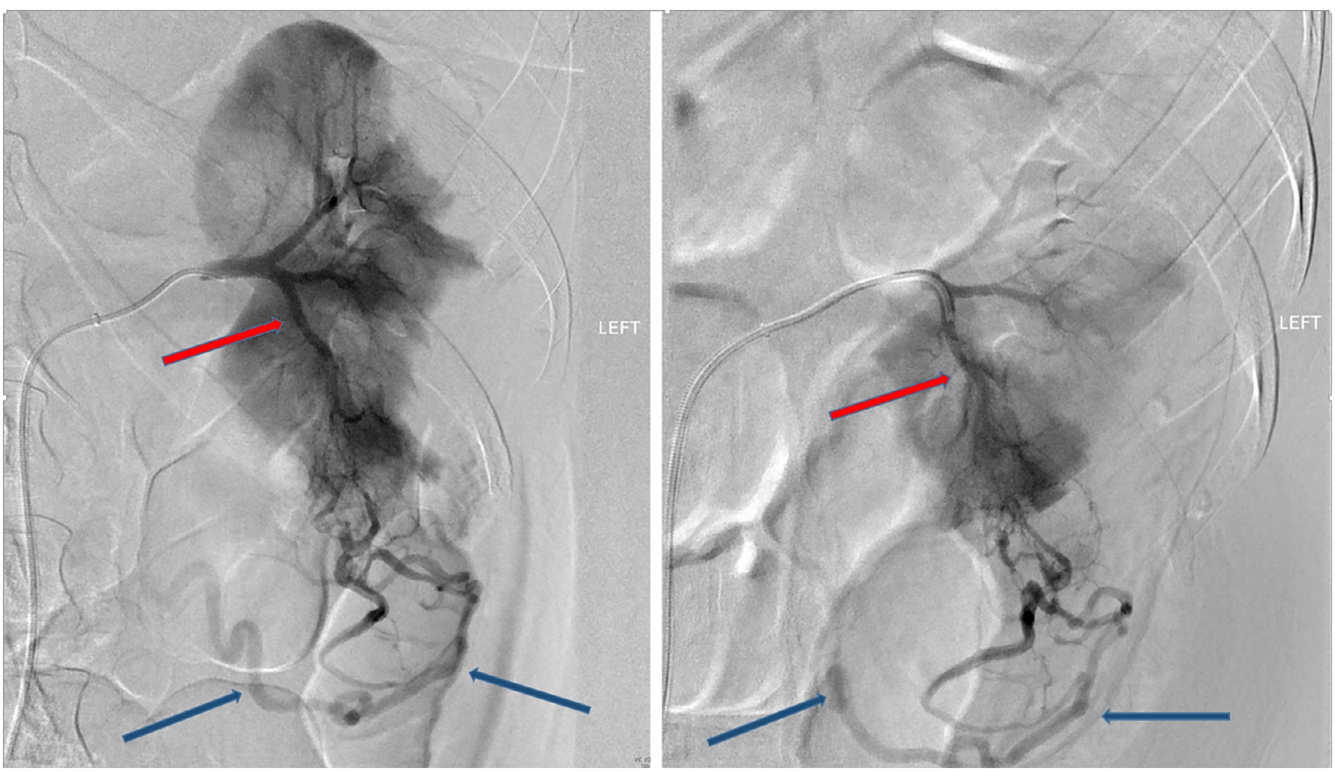

Fig. 2 – Selective angiography of the left renal artery and lower renal artery branch (red arrows), revealing early venous filling with rapid flow, draining toward the inferior vena cava (blue arrow) consistent with arteriovenous shunting.

Department with flank pain and anemia. Computed tomography (CT) revealed 2 large renal AMLs on the left, 1 arising from the upper pole (Fig. 1a) and another arising from the lower pole, which appeared to be the source of hemorrhage (Fig. 1b). The patient required blood transfusion and fluid resuscitation in the Emergency Department, prompting an urgent consult to Interventional Radiology for embolization.

Abdominal aortography demonstrated a single left renal artery with early branching. A lower pole branch of the renal artery and a small vessel arising from the aorta were found supplying the lower pole lesion (Fig. 1c).

Selective angiography of the left renal and lower branch artery revealed early venous filling, with rapid flow, draining toward the inferior vena cava consistent with arteriovenous (AV) shunting (Fig. 2). This finding precluded the use of embolization agents such as absolute alcohol and small particles. Given the urgency of controlling hemorrhage, proximal embolization was performed using large gelatin foam (Gelfoam)

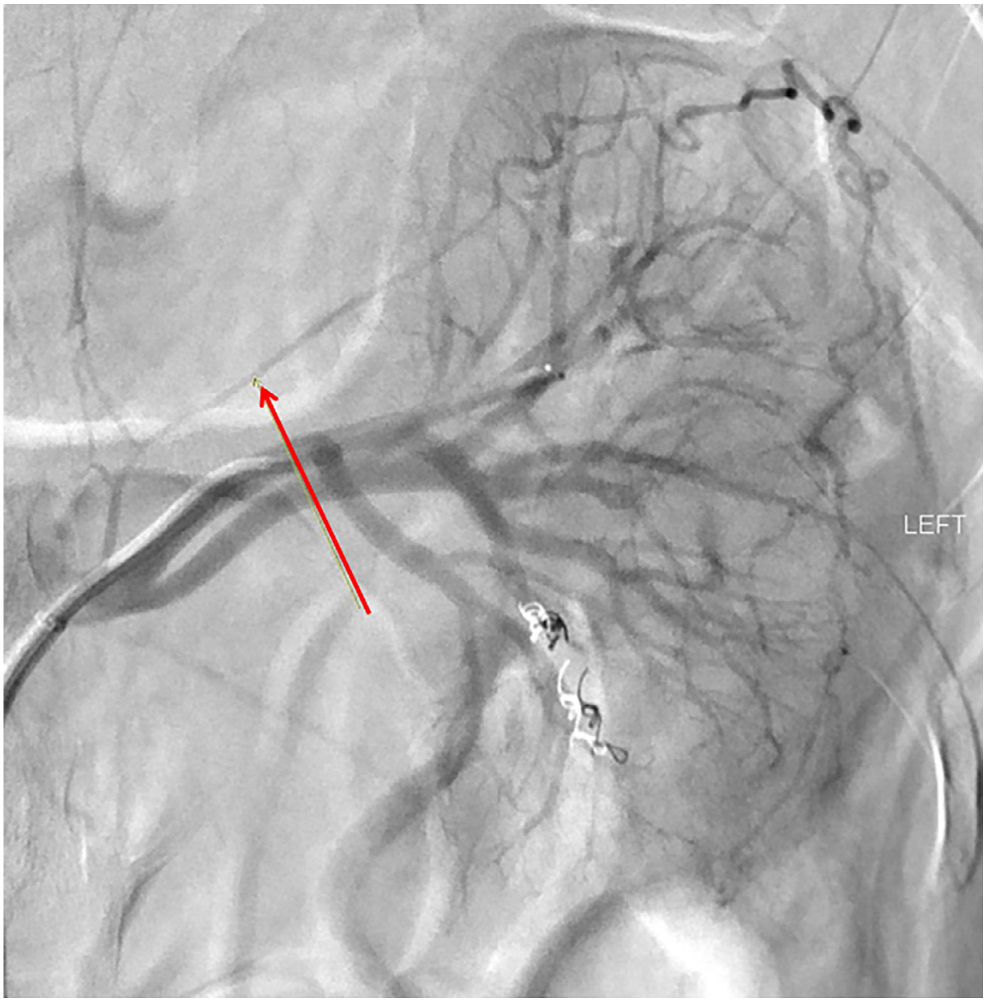

Fig. 3 – Completion angiography revealing a capsular artery circumscribing the left kidney, supplying the lower pole lesion (red arrow). This was selectively catheterized and embolized using a combination of large gelatin foam particles and metallic coils.

particles and metallic coils. No additional arterial supply to the lesion through the left renal arterial system was identified on completion angiography. There was no evidence of tumor blush noted during selective catheterization of the upper pole branch of the left renal artery.

Completion angiography revealed a capsular artery circumscribing the left kidney and supplying the lower pole AML. This was selectively catheterized and embolized using a combination of large Gelfoam particles and metallic coils (Fig. 3).

Completion aortography demonstrated a lumbar artery supplying a portion of the lower pole lesion. Selective catheterization was performed, and a combination of Gelfoam and metallic coils was again utilized to embolize the artery. Completion angiogram was performed which demonstrated satisfactory appearance (Fig. 4).

The patient remained hemodynamically stable, without significant decrease in hemoglobin following the procedure, but continued to have pain. Follow-up CT was performed 2 days later to evaluate the adequacy of embolization, which not unpredictably revealed residual vascularity within the lesion.

A second procedure was subsequently performed, which demonstrated reconstituted involvement of the lower pole branch of the left renal artery. The AV shunt now demonstrated markedly slower flow and later filling as a result of the proximal embolization during the first procedure, thus allowing the option of using ethylene vinyl alcohol copolymer dissolved in dimethyl sulfoxide (Onyx).

Embolization of the involved renal artery branches using high viscosity ethylene vinyl alcohol copolymer dissolved in dimethyl sulfoxide was then performed, demonstrating satisfactory results (Fig. 5a). Involvement of a new lumbar artery was noted on completion angiography, and again the same high viscosity agent was used to embolize with satisfactory results (Fig. 5b). The patient remained stable after the procedure and post discharge. Follow-up imaging showed interval resolution of the retroperitoneal hematoma without evidence of renal dysfunction.

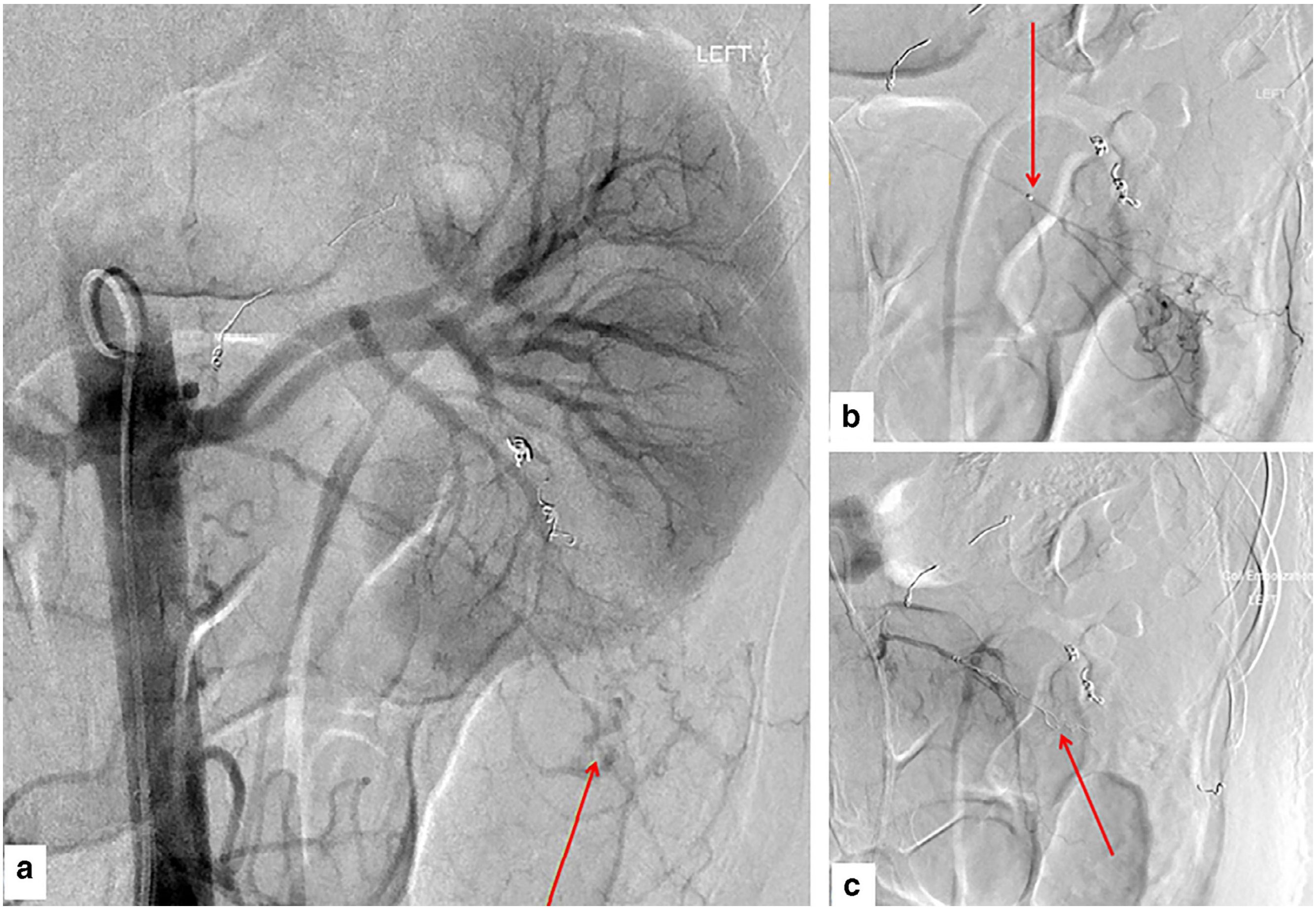

Fig. 4 – (a,b) Completion aortography demonstrating a lumbar artery branch supplying a portion of the lower pole lesion (red arrows). (c) Large gelatin foam and metallic coils were again utilized for embolization (red arrow).

Discussion

AML usually present as isolated tumors, but can be seen in up to 80% of patients with hereditary tuberous sclerosis [1,2]. The classic AML is composed of adipose tissue, spindle cells, and smooth muscle cells with increased vascularity and vessel wall thickening [1]. Sporadic AMLs have a female preponderance, occurring between the ages of 30 and 50 years, and are more often unilateral and larger in size [1].

The radiological appearance varies, adding challenge to differentiating these lesions from a renal cell carcinoma [2]. The classic CT appearance is a fat containing homogenous mass. Angiographic features include neovascularity, whorled appearance on venous phase, terminal aneurysms, and arterial "corkscrewing" [3,4]. AV shunting in association with AML is unusual [4–7]. In equivocal cases, its presence is sometimes used as a distinguishing feature of renal cell carcinoma [7].

The classic Lenk's triad of renal AML includes flank pain, a palpable tender mass, and signs of internal bleeding. Rupture can result in hypovolemic shock, and the risk of hemorrhage increases with tumor size [8]. Lesions larger than 4 cm are symptomatic in approximately 90% of cases, and carry and increased risk of hemorrhage, compelling intervention [3,8].

Management aim is to relieve symptoms and prevent or control hemorrhage, while preserving renal function [9]. Treatment options include arterial embolization, ablation therapies, or surgical excision [8,9]. In the setting of acute hemorrhage, embolization provides rapid hemodynamic stabilization, with maximal preservation of renal function, and the advantage of being minimally invasive [5,10,11]. Surgical resection is reserved when the diagnosis of renal cell carcinoma cannot be excluded, or when hemorrhage cannot be controlled using less invasive means [4,9].

AML can have complex vascular anatomy necessitating embolization of multiple branches [12]. Such cases can be technically challenging, with an increased risk of embolizing adjoining normal renal parenchyma, resulting in functional compromise [5,12]. Embolization options that have been successfully used to treat renal AML include polyvinyl alcohol foam powder with coils, polyvinyl alcohol with alcohol, alcohol with ethiodized oil, and sterile compressed sponge [13].

However, the use of particle- or liquid-based embolization materials is not suitable in the presence of AV shunt hemodynamics [13]. Gelatin foam with metallic coils has been demonstrated as a suitable option allowing embolization with reduced risk of nontarget embolization through the AV shunt [13,14]. A disadvantage to this approach is the high frequency

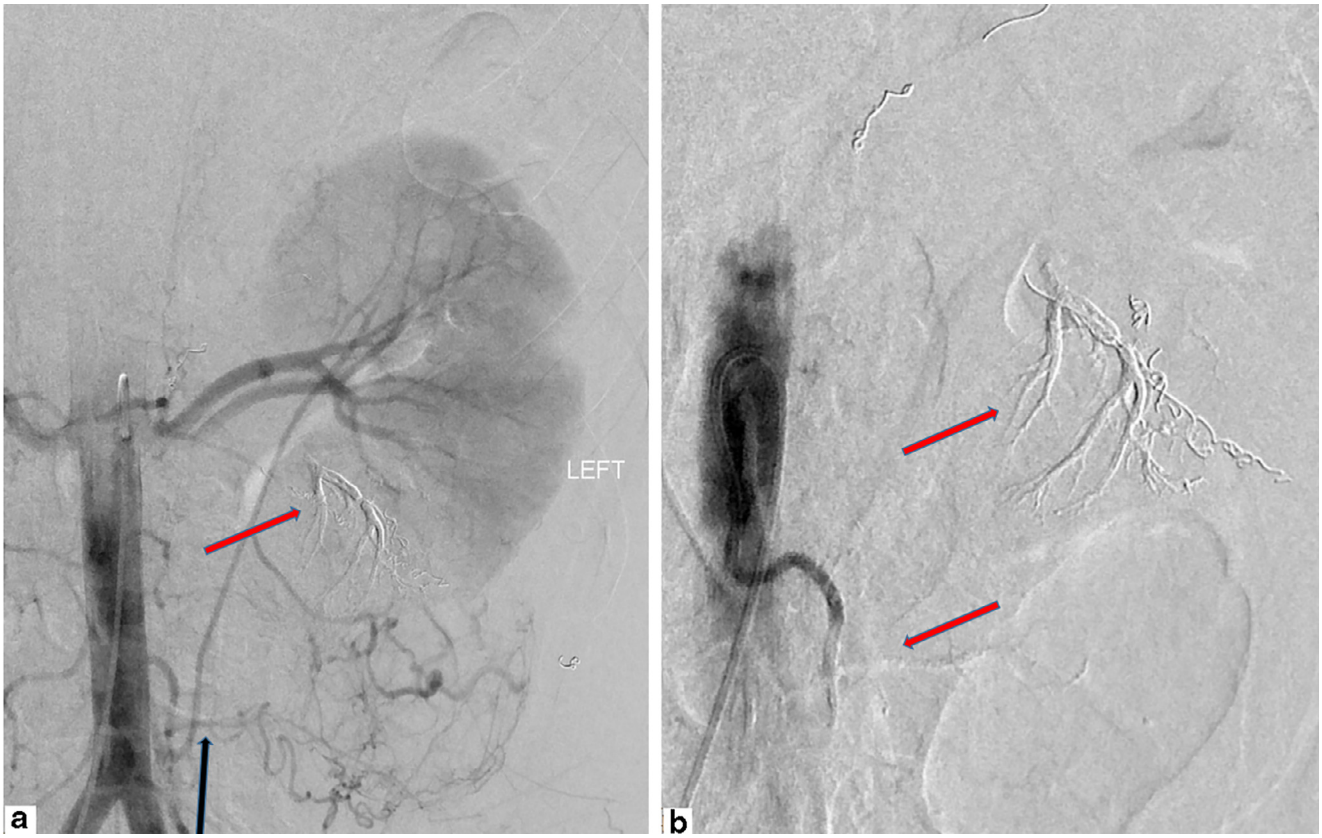

Fig. 5 – (a) Decreased flow in the arteriovenous shunt allowed the option of using high viscosity Onyx during the second procedure (red arrow). (b) Involvement of a new lumbar artery was noted (black arrow), and again Onyx was used with satisfactory results (red arrow).

of collateral vessel recruitment, which we encountered in this case [13]. We employed a 2-step approach in which proximal embolization using large gelatin foam and metallic coils was performed, resulting in stabilization of hemorrhage and reduction in AV shunt flow. This allowed for subsequent selective embolization using high viscosity liquid for definitive treatment.

Conclusion

Transcatheter arterial embolization is an effective therapy for the treatment of spontaneous hemorrhage in AML. In unusual cases where AV shunting is present, the use of traditional embolic agents is precluded. In this case, proximal embolization provided hemodynamic stability and reduced flow through the AV shunt, allowing for more definitive treatment using a high viscosity liquid embolic agent.

Competing Interests

None.

Supplementary materials

Supplementary material associated with this article can be found, in the online version, at doi:10.1016/j.radcr.2018.07.004.

REFERENCES

[1] Martignoni G, Pea M, Reghellin D, Zamboni G, Bonetti F. PEComas: the past, the present and the future. Adv Anat Pathol 2007;14(1):36–41 doi:0.1097/PAP.0b013e31802e0dc4.

[2] Siegel CL, Middleton WD, Teefey SA. McLennan BL angiomyolipoma and renal cell carcinoma: US differentiation. Radiology 1996;198:789–93.

[3] Halpenny D, Snow A, McNeill G, Torreggiani WC. The radiological diagnosis and treatment of renal angiomyolipoma—current status. Clin Radiol 2010;65:99–108. doi:10.1016/j.crad.2009.09.014.

[4] Kehagias D, Mourikis D, Kousaris M, Chatziioannou A, Vlahos L. Management of renal angiomyolipoma by selective arterial embolization. Urol Int 1998;60:113–17.

[5] Mues AC, Palacios JM, Haramis G, Casazza C, Badani K, Gupta M, et al. Contemporary experience in the management of angiomyolipoma. J Endourol 2010;24(11):1883–6. doi:10.1089/end.2010.0223.

[6] Barzilai IM, Braden GL, Ford LD, Goodman LH, Delima RJ, Germain MJ, et al. Renal angiomyolipoma with arteriovenous shunting. J Urol 1987;137(3):483–4.
[7] Danneels F, Meeus L, Casselman JW, Pattyn G, Vandevoorde P, Steyaert L, et al. Giant angiomyolipoma with atypical arteriovenous shunting: CT and angiographic studies. J. Belge Radiol 1990;73(2):135.
[8] Yamakado K, Tanaka N, Nakagawa T, Kobayashi S, Yanagawa M, Takeda K. Renal angiomyolipoma: relationships between tumor size, aneurysm formation, and rupture. Radiology 2002;225:78–82. doi:10.1148/radiol.2251011477.
[9] Urciuoli P, D'Orazi V, Livadoti G, Foresi E, Panunzi A, Anichini S, et al. Treatment of renal angiomyolipoma: surgery versus angioembolization. G Chir 2013;34:326–31; Sivalingam S, Nakada SY. Contemporary minimally invasive treatment options for renal angiomyolipomas. Curr Urol Rep 2013;14(2):147–53 Apr. doi:10.1007/s11934-013-0311-3.
[10] Patatas K, Robinson GJ, Ettles DF, Lakshminarayan R. Patterns of renal angiomyolipoma regression post-embolisation on medium to long-term follow-up. Br J Radiol 2013;86(1024):20120633. doi:10.1259/bjr.20120633.
[11] Villalta JD, Sorensen MD, Durack JC, Kerlan RK, Stoller ML. Selective arterial embolization of angiomyolipomas: a comparison of smaller and larger embolic agents. J Urol 2011;186(3):921–7 Sep. doi:10.1016/j.juro.2011.04.082.
[12] Sivalingam S, Nakada SY. Contemporary minimally invasive treatment options for renal angiomyolipomas. Curr Urol Rep 2013;14(2):147–53 Apr. doi:10.1007/s11934-013-0311-3.
[13] Kikuchi N, Kuwatsuru R, Kyogoku S, Shiraishi A, Okada S, Tsuge D, et al. Embolization of spontaneous intratumoral hemorrhage with the hemodynamic characteristics of arteriovenous fistula in renal angiomyolipoma. Case Rep Nephrol Dial 2015;5(1):54–9. doi:10.1159/000371844.
[14] Nassiri N, Ali H, Carroccio A, Rosen RJ. Transarterial treatment of congenital renal arteriovenous fistulas. J Vasc Surg 2013;58(5):1310–15.