Embolization of a sporadic bleeding renal angiomyolipoma supplied by an aberrant renal artery.

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ABSTRACT: Spontaneous bleeding of a renal angiomyolipoma (AML) is a life threatening condition that necessitates prompt treatment. Herein we describe the first case of a spontaneously ruptured renal AML supplied solely by an aberrant renal artery that was successfully treated by selective transcatherer embolization.

Key Words: Renal angiomyolipoma, Bleeding, Embolization.

INTRODUCTION

Angiomyolipoma (AML) is a benign renal tumor composed by various proportions of mature adipose tissue, abnormal thick-walled blood vessels and smooth muscle. Renal AMLs are prone to spontaneous bleeding due to dysplastic aneurysmal vessels. Selective transcatheter arterial embolotherapy (TAE) of ruptured AML is considered nowadays as the treatment of choice [1-3]. Herein we present the first case of a spontaneously ruptured sporadic renal AML supplied exclusively by an aberrant renal artery that was successfully treated with TAE.

CASE REPORT

A 52-year-old woman with free medical history, presented with acute right flank pain. She was in good physical condition and hemodynamically stable. Blood and urine examination were unremarkable. Abdominal ultrasound demonstrated a right perirenal collection with hyperechoic components and a hyperechoic hypervascular lesion of the lower pole of the right kidney.

Due to suspicion of a ruptured renal tumor, the patient was immediately transferred for abdominal computed tomography (CT). Unenhanced and contrast-enhanced (in arterial and delayed phase) images demonstrated a non-enhancing hyperdense perirenal collection and a renal vascular lipomatous tumor (Figure 1A). The diagnosis of a spontaneously ruptured renal AML was established.

Further evaluation with digital subtraction angiography demonstrated an AML of the lower pole of the right kidney. A right aberrant renal artery supplying the lower pole of the right kidney was faintly demonstrated (Figure 1B). Selective angiography of the main and aberrant renal arteries demonstrated that the AML was solely supplied by the aberrant renal artery (Figure 1C). Taking under consideration these findings and after discussion with the patient, embolization of the aberrant renal artery was decided. Polyvinyl alcohol particles (PVA, 300-500 μm) were initially used in order to occlude the tumor microcirculation and then three platinum coils (5mm/35mm) were used in order to occlude the inflow artery (Figure 1D-E).

Post-embolization period was uneventful, without signs or symptoms of post-embolization syndrome and the patient was discharged from the hospital after 4 days. CT 1 and 12 months post treatment demonstrated significant decrease of the lesion size, with no contrast enhancement (Figure 1F-G).

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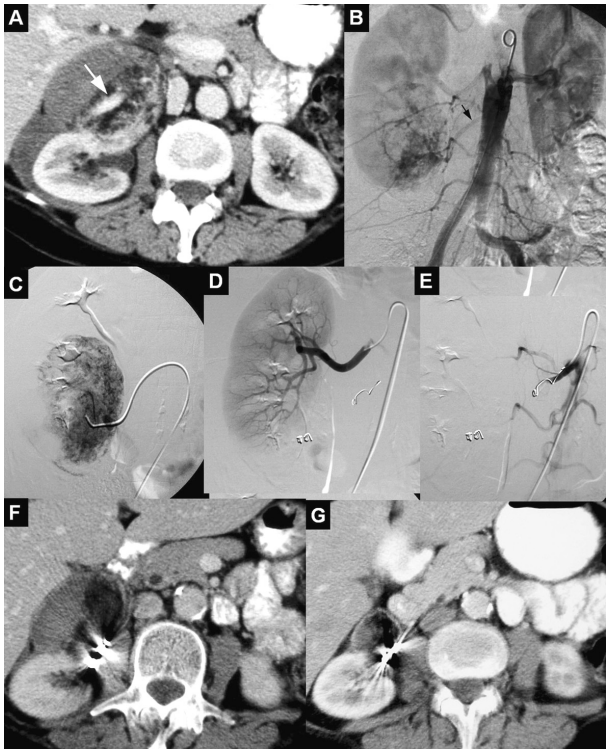

Figure 1. A. Contrast enhanced CT in arterial phase, demonstrating a bleeding renal AML with perirenal hematoma. Note the dilated vascular components of the AML (arrow).
B. Aortography, demonstrating the AML of the lower pole of the right kidney, with the abnormal vascularity within the lesion. An aberrant artery is visualized, supplying the lower pole of the kidney (arrow).
C. Selective angiography of the aberrant renal artery, demonstrating the dysmorphic tortuous vasculature of the lesion and the aneurysms within the AML.
D-E. Selective angiography of the main (D) and aberrant renal artery (E) after embolization demonstrating complete occlusion of the vasculature of the lesion.
F-G. Contrast enhanced CT scans after 1 month (F) and 12 months (G), demonstrating gradual absorption of the subcapsular hematoma and decrease of the lesion size (>80%), while the right kidney is demonstrated morphologically and functionally intact.

DISCUSSION

Renal AMLs are slow growing benign renal tumors found either isolated or in association with tuberous sclerosis[1,2,4]. Spontaneous bleeding of a renal AML is the most serious and potentially life-threatening complication. Treatment of renal AMLs depends on the tumor size and symptoms. Asymptomatic AMLs < 4cm should be managed conservatively. Interventional treatment should be performed in bleeding AMLs, AMLs with a history of hemorrhage, AMLs causing pain and hematuria and in lesions > 4cm because they have an increase risk of spontaneous bleeding[1]. TAE is considered nowadays as the treatment of choice, with preservation of as much functioning renal tissue as possible, with reduced morbidity and mortality compared to total nephrectomy or nephron-sparing surgery[1,2,4-8]. A variety of embolic materials has been used for TAE of renal AMLs targeting in occlusion of tumor circulation, control of hemorrhage and reduction of future bleeding risk. PVA are widely used in order to occlude the tumor microcirculation. PVA may be used alone or in combination with coils that additionally occlude the inflow artery. Coils may also be used alone[1,8,9]. The disadvantage of coiling the inflow artery is the occlusion of access for future re-embolization of the tumor in case or re-canalization[1,2]. On the other hand, when PVA or other liquid material are used alone, occlusion of the distal vascular bed results in increased vascular resistance and elevated pressure within the patent proximal feeding arteries, increasing the risk of periprocedural aneurysm rupture and the long-term recurrence rate[1,2,7]. Other embolic materials used for renal AMLs are absolute alcohol with or without iodized oil[2,6-8], combination of PVA with gelatin sponges (Gelfoam)[8] and onyx (ethylene vinyl alcohol copolymer)[5].

Extra renal arteries (accessory and aberrant renal arteries) are detected angiographically in 24% of the patients[10]. Visualization of these variations is very important in cases of urological and vascular operations, renal transplantation and interventional radiological procedures. In our case, selective angiography of both main and aberrant renal arteries was crucial for the decision of the treatment plan.

In conclusion, we present the first case of a ruptured renal AML supplied exclusively by an aberrant renal artery that was successfully treated by TAE. Although not reported in the literature, we believe that it

is of major importance to exclude any arterial supply of a ruptured AML by extra renal arteries (which are so common) in order to avoid incomplete TAE treatment with failure of bleeding control and possible future relapse.

Abbreviations:
Angiomyolipoma (AML), transcatheter arterial embolotherapy (TAE), polyvinyl alcohol particles (PVA), computed tomography (CT).

Εμβολισμός σποραδικού νεφρικού αγγειομυολιπώματος με αιμάτωση από επικουρική νεφρική αρτηρία.

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ΠΕΡΙΛΗΨΗ: Η αυτόματη αιμορραγία ενός νεφρικού αγγειομυολιπώματος αποτελεί μια κατάσταση απειλητική για τη ζωή που χρήζει άμεσης θεραπείας. Παρουσιάζουμε την πρώτη περίπτωση αυτόματης ρήξης νεφρικού αγγειομυολιπώματος με αποκλειστική αιμάτωση από επικουρική νεφρική αρτηρία που αντιμετωπίστηκε επιτυχώς με εκλεκτικό εμβολισμό.

Λέξεις Κλειδιά: Αγγειομυολίπωμα νεφρού, Αιμορραγία, Εμβολισμός.

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