

## Selective angioembolization as a management strategy for angiomyolipoma causing urinary tract obstruction

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### ARTICLE INFO

#### Keywords:

Angiomyolipoma  
Selective angioembolization  
Hydronephrosis

### ABSTRACT

Angiomyolipoma (AML) is a benign renal tumor usually found incidentally. Rarely, AML can present with renal colic due to urinary tract obstruction. Prior cases of obstructing AML have been presented and managed successfully with surgical removal. Selective angioembolization has emerged as an alternative management strategy for AML, but no documented cases have been presented for this strategy in the setting of obstruction. Here, we present a case of obstructing AML treated with selective angioembolization with subsequent resolution of obstruction.

### 1. Introduction

Renal angiomyolipoma (AML) is a benign renal tumor composed of adipose tissue, dystrophic blood vessels, and smooth muscle in varying proportions.<sup>1</sup> It most often occurs sporadically but also has a known association with Tuberous Sclerosis Complex (TSC) and pulmonary lymphangiomatosis.<sup>1</sup> Prevalence ranges from 0.2 to 0.6%, most commonly occurring in middle-aged women.<sup>1</sup> Around 80% of AMLs are diagnosed incidentally, but up to 10% present with retroperitoneal bleeding.<sup>1</sup> Treatment options include embolization, partial nephrectomy, nephrectomy, ablation, mTOR inhibitors (such as everolimus), and active surveillance.<sup>2</sup> Angioembolization has been adopted as an emerging first-line treatment with high efficacy and low risk of complications. Only 2 cases have been reported in the literature of an AML causing urinary flow obstruction, which were treated with simple and partial nephrectomy.<sup>3,4</sup> Here we present a case of an obstructing AML successfully managed with angioembolization.

### 2. Case presentation

Our patient is a 68 year old man with a known right AML which over several years increased from 3.5 to 5.5 cm. Despite the size, the tumor was managed with surveillance due to its perihilar location. He

presented with worsening right-sided flank pain progressing over several days. Evaluation revealed normal vital signs, a hemoglobin of 15.9 g/dL, and creatinine level of 1.46 mg/dL from a normal baseline. With clinical suspicion for an obstructing stone CT scan was performed revealing new moderate right hydronephrosis without stone. The obstruction was determined to be due to increased size of the known AML (Fig. 1-A,B) now compressing and displacing the proximal right ureter leading to obstruction. Following discussion, the patient elected for placement of right percutaneous nephrostomy tube (PCN) to relieve the obstruction. In the same procedure a selective angioembolization of the AML was to be performed with the understanding that a more invasive approach may be needed should the AML not respond to the angioembolization. Right PCN was placed and antegrade nephrostogram confirmed obstruction with diminished contrast seen traversing the proximal ureter (Fig. 1-C). Arteriography revealed vascular supply to the AML as a subsegmental branch of the right inferior segmental artery. Superselective angioembolization was performed using ethanol and lipiodol. Repeat angiogram showed no residual vascular flow to the AML. The patient was discharged home the following morning after his creatinine normalized to 1.11 mg/dL. The PCN remained in place for 6 weeks. He returned for repeat antegrade nephrostogram which revealed brisk contrast passage from the renal collecting system through the proximal ureter to the bladder (Fig. 1-D). CT scan at this time

*Abbreviations:* TSC, Tuberous Sclerosis Complex; AML, Angiomyolipoma; PCN, Percutaneous Nephrostomy.

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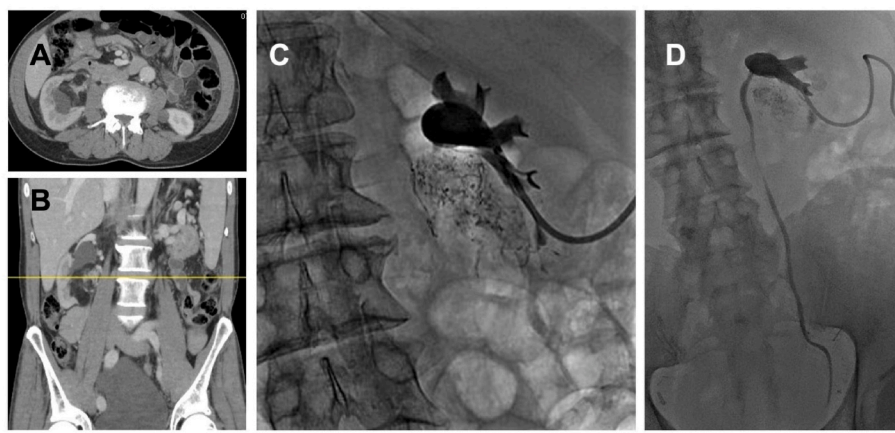
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<https://doi.org/10.1016/j.eucr.2022.102240>

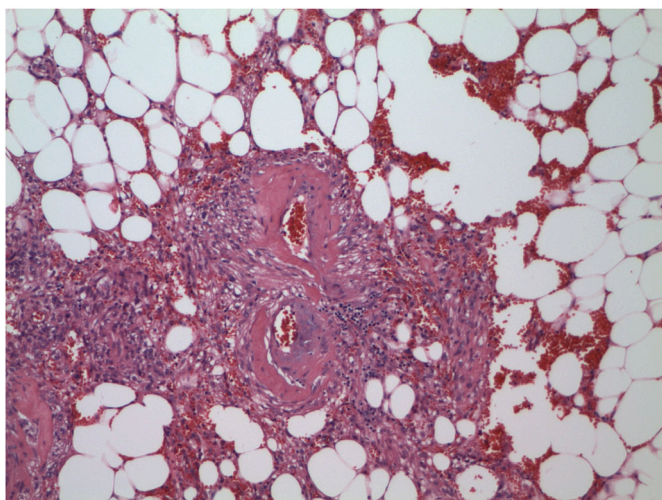
Received 18 August 2022; Received in revised form 19 September 2022; Accepted 23 September 2022

Available online 24 September 2022

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**Fig. 1.** (A&B) Right AML of the lower pole leading to obstruction of the renal pelvis. (C) Right antegrade nephrostogram at initial presentation showing opacification obstructed right renal collecting system without any contrast noted distal to the renal pelvis. (D) Antegrade nephrostogram 6 weeks following embolization showing free flow of contrast from the renal pelvis to the ureter with no obstruction.



**Fig. 2.** Microscopic image of typical angiomyolipoma (not this patient). The three components are present with central dysmorphic blood vessels from which there is radiating smooth muscle and surrounding adipose tissue. H&E 10x

demonstrated decreased size to 3.5 cm. The PCN was capped for three weeks prior to removal and the patient remained asymptomatic. His creatinine further improved to 0.78 mg/dL and he was without pain. The tube was then removed, and the patient continues to do well now 2 months following PCN removal.

### 3. Discussion

AML is a benign mesenchymal neoplasm consisting of varying amounts of adipose tissue, dysmorphic blood vessels, and smooth muscle (Fig. 2).<sup>1</sup> The tumor is considered a member of the perivascular epithelioid cell (PEC) family. AMLs occur sporadically 80% of the time and are associated with TSC in up to 20% of the time.<sup>1</sup> Our case is a representation of the former. AML is more likely to present as a larger mass, multifocal, or bilateral when associated with TSC.<sup>1</sup> Multiple variants based on histological subtype have been described in the literature, and some such as the epithelioid variant express more aggressive behavior with potential for metastases and mortality.<sup>5</sup> AML is most commonly asymptomatic and discovered incidentally. AML can present with various symptoms including hematuria, anemia, urinary infection, renal failure, and palpable flank mass. The most significant clinical

concern is life-threatening retroperitoneal hemorrhage most often occurring in tumors >4 cm. Our patient was symptomatic and presented with flank pain secondary to the AML causing obstruction and hydronephrosis, which is a very rare presentation.

AML is identified using various imaging modalities and sometimes requires biopsy depending on the radiologic classification. CT and MRI have equal accuracy in AML detection and diagnosis.<sup>5</sup> In lesions with high fat content, the diagnosis of AML can accurately be made with imaging alone. However, fat-poor lesions can be difficult to diagnose and to differentiate from other parenchymal renal masses like renal cell carcinoma. In these select cases, percutaneous biopsy can be utilized to achieve accurate diagnosis. Ultrasound may also be used as an imaging modality but lacks sensitivity in defining small renal masses.<sup>5</sup> Prior studies have also identified fat content as a predictor of size reduction following SAE. Specifically, lesions with >50% fat are much more likely to experience size reduction than lesions with <50%.<sup>5</sup>

Many treatment modalities for AML have been utilized including nephrectomy, partial nephrectomy, selective angioembolization, and ablation.<sup>2</sup> The mTOR inhibitors have also been used in cases of TSC or when angioembolization or nephrectomy is not feasible, especially when multifocal masses are present and would result in increased nephron loss with SAE.<sup>2</sup> Masses over 4 cm or those that are symptomatic should be treated.<sup>2</sup> Angioembolization is a first-line treatment, especially in the setting of bleeding and is also typically utilized for tumors >4 cm, or symptomatic lesions to reduce size, symptoms, and risk of hemorrhage. Compared to surgical intervention, angioembolization is associated with less postoperative complications and better preservation of renal function.<sup>1</sup> However, when compared to nephrectomy angioembolization has higher rates of recurrence and need for additional procedures.<sup>1</sup>

Two prior cases of obstructing AML have been reported.<sup>3,4</sup> Each were treated surgically with excision. This report describes the first case of obstructing AML treated with angioembolization. It should be considered that it may take weeks for a noticeable size decrease to occur following embolization. Such patients will need ureteral stents or PCN to relieve the obstruction and protect the kidney while treatment takes effect. Angioembolization does not come without risks. Prior embolization may make partial nephrectomy increasingly difficult, especially with a perihilar mass. Although in our patient treatment was successful, patients should receive thorough counseling on benefits and risks of embolizing an obstructive AML, especially on the risk of kidney loss if surgery were to be needed.

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