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*CASE REPORT*

# Treatment of fat-poor renal angiomyolipoma with ectopic blood supply by fluorescent laparoscopy: A case report and review of literature

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## Abstract

*BACKGROUND*

Renal angiomyolipoma and renal cell carcinoma are the most common benign and malignant tumors of the kidney respectively, and the preoperative differential diagnosis is crucial due to the wide difference in treatment methods. Fat-poor renal angiomyolipoma is a relatively rare type of in renal angiomyolipoma. Its fat imaging features are not obvious, and it is easily misdiagnosed as renal cell carcinoma.

*CASE SUMMARY*

We report the case of a 41-year-old man who complained of osphyalgia. Subsequent abdominal computed tomography scans revealed that a heterogeneous mass was seen in the lower pole of the right kidney, with the size of about 53 mm × 47 mm. And showed two right renal arteries, with the mass supplied by an ectopic vessel from the abdominal aorta. Fluorescent laparoscopic blockade of the right renal heterotopic artery and partial nephrectomy was performed. Based on histological and immunohistochemical findings, the tumor was diagnosed as fat-poor renal angiomyolipoma.

*CONCLUSION*

The use of fluorescent laparoscopy can effectively help intraoperative management, and the fluorescence pattern provided by intravenous indocyanine green can help suggest the final diagnosis, effectively guide the surgical decision-making, and avoid preoperative imaging diagnosis leading to nephrectomy for benign renal tumors, through fluorescent navigation of tumor supply vessel precise block, minimize the loss of renal function.

**Key Words:** Renal angiomyolipoma; Renal cell carcinoma; Ectopic blood supply; luorescent laparoscopic; Partial nephrectom; Case report



**Core Tip:** We used the PINPOINT fluorescent laparoscopic system intraoperatively. The fluorescence pattern provided by intravenous indocyanine green helped to suggest the final diagnosis and effectively guided surgical decision making, avoiding nephrectomy for benign renal tumors. In the fluorescence mode, we only blocked the ectopic blood vessels supplying the tumor, thus minimizing the loss of renal function.

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## INTRODUCTION

Kidney tumors are mostly found incidentally by B-ultrasound or computed tomography (CT). Renal angiomyolipoma (RAML) is the most common benign tumor of the kidney, RAML is usually composed of smooth muscle, blood vessels, and mature adipose tissue. Most cases, the typical RAML can be achieved by color Doppler ultrasound (Dopplerul-trasound), CT or magnetic resonance imaging (MRI) to diagnose. However, approximately 4%–5% of fat-poor RAML could not be diagnosed by the above examinations[1]. Fat-poor RAML is a relatively rare type of renal angiomyolipoma [2], and the proportion of fat components in its tumor is < 20%, which is easily misdiagnosed as renal cell carcinoma[3]. At present, only a few cases have been reported about fat-poor RAML, however, fat-poor RAML with independent branches of the abdominal aorta has not been reported. We performed a partial nephrectomy with the aid of fluorescent laparoscopy.

## CASE PRESENTATION

### *Chief complaints*

A 41-year-old man patient, who complained of osphyalgia in the right waist for more than 1 month, was admitted to the hospital in June 2023.

### *History of present illness*

The patient complained of right low back pain discomfort for more than 1 month with no apparent cause. He was unconcerned by it at the time. The patient reported no nausea, vomiting, numbness in the extremities or trunk, muscle weakness, or urinary disruption. However, the symptoms were repeated, and then he came to the hospital for further examination.

### *History of past illness*

There is no obvious history of past illness related to this disease.

### *Personal and family history*

There is no obvious personal or family history related to this disease.

### *Physical examination*

On admission to the hospital, he was conscious; his vital signs were stable, and no enlarged lymph nodes were discovered in the neck or behind the ears. Both muscle strength and muscle tension of the extremities were normal, and voluntary activities were normal. There was no significant palpable mass in the abdomen.

### *Laboratory examinations*

Routine blood tests, blood biochemistry, tumor markers, immune markers, infection markers, routine urine tests and routine stool tests showed no significant abnormalities.

### *Imaging examinations*

B-ultrasound showed a right renal mass with a few high echoic areas, considered renal cell carcinoma, subsequent abdominal contrast-enhanced CT revealed that a heterogeneous mass was seen in the lower pole of the right kidney, with

the size of about 53 mm × 47 mm, which had moderately uneven enhancement (Figure 1). There was significant enhancement in the arterial phase and decreased density in the venous and excretory post-contrast phases. Of special interest was the mass with ectopic kidney arterial blood supply that was enhanced on CT (Figure 2A and B). The ectopic artery was emitted from the abdominal aorta, below the renal arteries, no significant stenosis and extruded of the renal artery.

## FINAL DIAGNOSIS

Right kidney tumor.

## TREATMENT

Based on all the examination results, we initially considered the mass as renal cell carcinoma, so the patient requested a radical nephrectomy.

The right kidney tumor was explored under laparoscopy, and radical nephrectomy would be performed if necessary. We used the PINPOINT fluorescent laparoscopic system (Novadaq Technologies, Mississauga, ON Canada) intraoperatively. An 2 mL of 2.5 mg/mL indocyanine green (ICG) (IC-Green, Akorn Pharmaceuticals, Lake Forest, IL, United States) solution was given intravenously for renal angiography. This allowed us to successfully identify the ectopic arterial vessels from the abdominal aorta, and clearly show that it supplied the middle and lower parts of the blood to the right kidney (Figure 3A and B). Also in the fluorescence mode, the renal tumor had almost the same color as the surrounding normal tissue, and the border is hard to recognize in the light of green or black and white (Figure 4A-D).

Our previous study[4] showed that renal cell carcinoma looked dark in the fluorescence mode. While other kidney tumors, such as renal angiomyolipoma, looked as bright as the normal renal tissues. Then, with the consent of the patient's family members, we performed a partial nephrectomy blocking the right renal heterotopic artery using fluorescent laparoscopy, the tumor was removed successfully and intact (Figure 5A and B). The time of renal artery ischemia during right partial nephrectomy is 35 minutes.

Histopathological images showed that the tumors was mainly composed of spindle cells and fat spindle cells with moderate cell density and a strip-like arrangement. The internal morphology of the tumor is diverse, with scattered thick-walled blood vessels and a small amount of mature adipose tissue, and a diffuse distribution of tumor cells between the blood vessels, closely related to the blood vessels and growing around the blood vessels. Immunohistochemical images showed: HMB 45 (+), SMA (+), C34 (vascular endothelial cells +), vim (+), S100 (adipocyte +), Ki-67 (+ 2%). The histological and immunohistochemical findings confirmed that it was a fat-poor RAML (Figure 6A-D). The fluorescence pattern provided by intravenous ICG can help suggest the final diagnosis, effectively guide the surgical decision-making, and avoid preoperative imaging diagnosis leading to nephrectomy for benign renal tumors.

## OUTCOME AND FOLLOW-UP

The patient was followed up for 12 months postoperatively, and no recurrence and metastasis were found. The renal function was not significantly increased than before surgery and was within the reference range.

## DISCUSSION

Due to advancements in modern imaging during the last years, nowadays more than 70% of kidney tumours are detected incidentally[5]. Renal cell carcinoma (RCC) is a common sporadic renal tumor that generally requires surgical resection. About 20% of solid renal tumors are benign, and RAML is more common[6]. RAML is mainly composed of mature spindle smooth muscle cells, deformed blood vessels and fat in different proportions. The proportion of various components varies greatly in different cases. Typical RAML often contains visible fat, while RCC has rare fat components [7], which can identify typical RAML and RCC. However, approximately 4%-5% of fat-poor RAML cannot diagnose[1] by the detection techniques described above. It is difficult to distinguish between atypical or fat-poor RAML and RCC on imaging, which easily leads to benign RAML being misdiagnosed as malignant RCC.

RAML is a relatively rare tumor with an incidence of less than 0.2%[8] and is the most common benign renal tumor. The disease was first reported by Fischer in 1911 and named[9] by Morgan in 1951. It was previously considered a hamartoma, but recent evidence suggests that RAML is a monoclonal rather than a tumor of polyclonal origin. Now, RAML is thought to originate from perivascular epithelioid cells of the neural crest and belongs to a member of the family of "perivascular epithelioid cell tumors". Currently, RAML is further classified into different tumor subtypes, each with its own unique pathological features, imaging features, and clinical manifestations[2].

Incidental RAML occurs in middle-aged women aged 40-60 years old, the ratio between women and men is about 4: 1, usually in one side of the kidney, Most asymptomatic during physical examination[1]. RAML may be associated by tuberous sclerosis (TSC), which tends to be multifocal and larger. Clinically, TSC occurs in approximately 20%-30% of

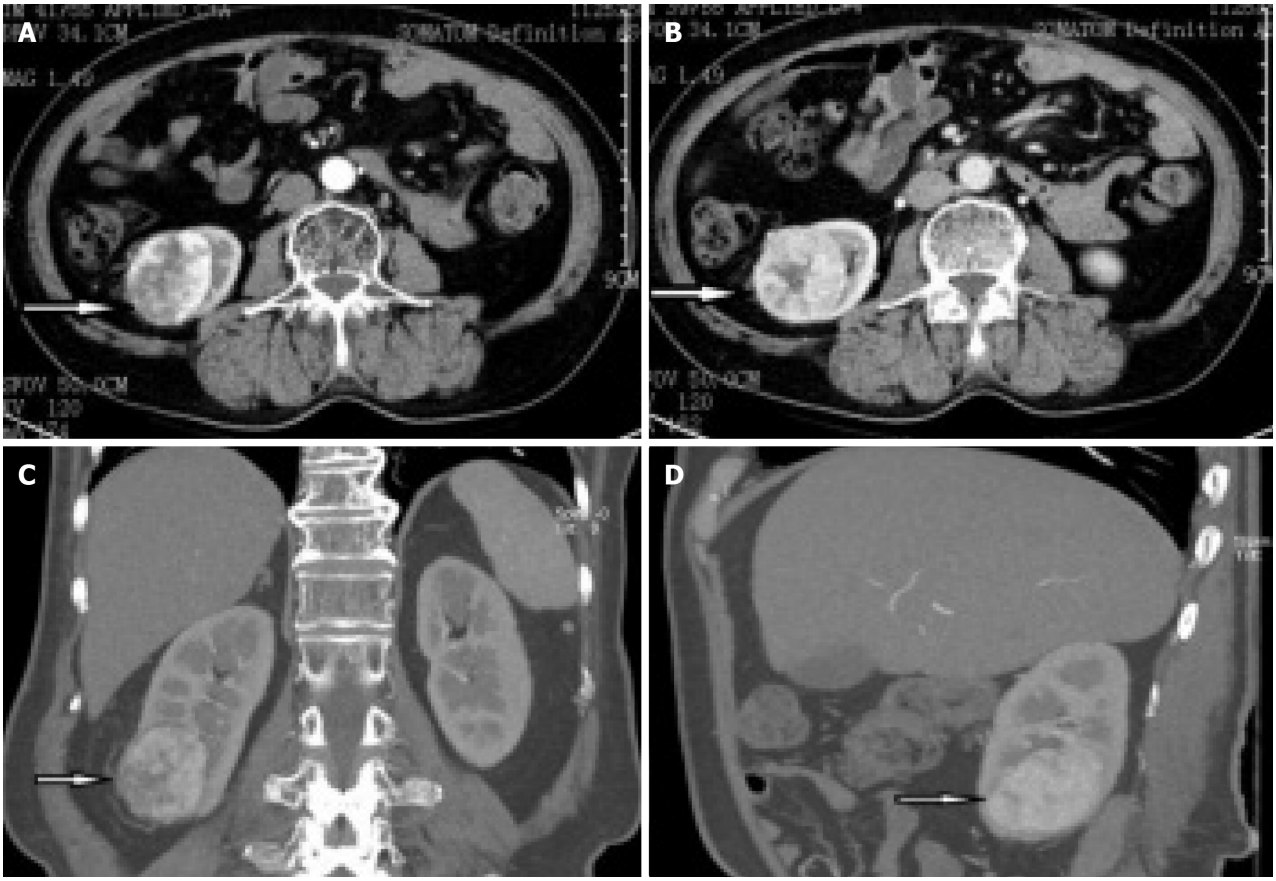


**Figure 1 Abdominal contrast-enhanced computed tomography scans revealed that a heterogeneous mass was seen in the lower pole of the right kidney, with the size of about 53 mm × 47 mm.** A: Arterial phase; B: Venous phase; C: Coronal section; D: Median sagittal section.

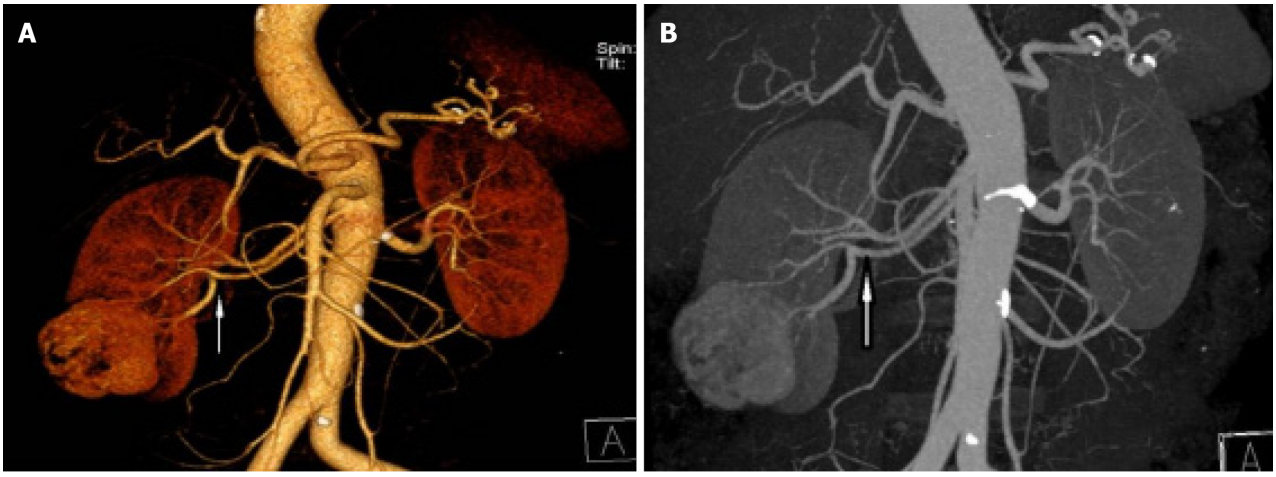


**Figure 2 A mass showing ectopic renal artery blood supply on contrast-enhanced computed tomography.** A and B: The ectopic artery flows from the abdominal aorta below the renal artery, supplying blood to the middle and lower pole part of the right kidney and the tumor.

patients with RAML, and approximately 50% of TSC patients develop RAML[1].

Fat-poor RAML is a relatively rare type of RAML[2]. The proportion of fat components in its tumors is < 20%, accounting for about 4.5% of all RAML. Its fat imaging features are not obvious, and it is easily misdiagnosed as renal cell carcinoma[10]. Most of the patients do not have lumbago. It is found in physical examination that its main components are smooth muscle and blood vessels, with very little fat composition, which is difficult to distinguish from renal cancer (especially clear cell carcinoma) in ordinary CT scan. Therefore, preoperative diagnosis of RAML is extremely important.

This patient was a 41-year-old male with B-ultrasound and enhanced CT both suggestive of right kidney tumor, considering malignancy, CT scans revealed that a heterogeneous mass was seen in the lower pole of the right kidney, with the size of about 53 mm × 47 mm. There was significant enhancement in the arterial phase and decreased density in the venous and excretory post-contrast phases. While showing no significant fat imaging. Of special interest was the mass

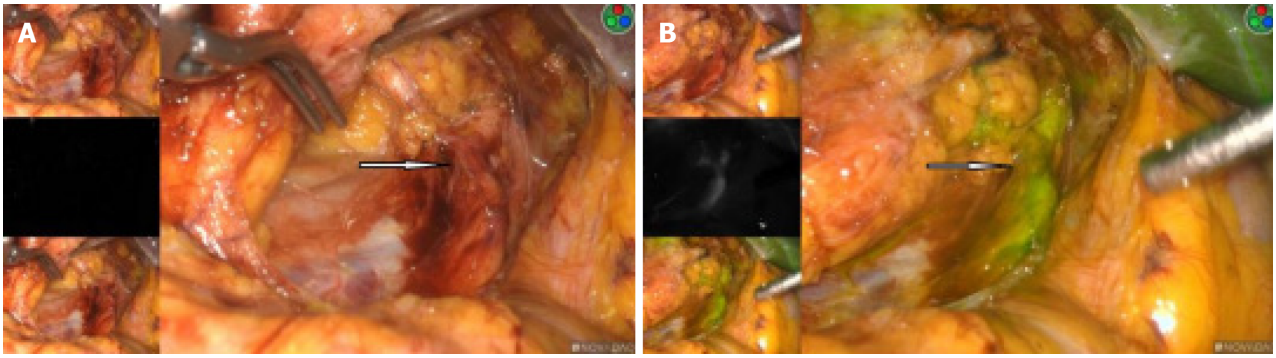


**Figure 3 The fluorescence mode clearly indicates that the ectopic artery provided blood to the middle and lower half of the right kidney.**
A: Artery showing an ectopic right kidney in the white-light mode; B: Artery showing an ectopic right kidney in the fluorescent mode.

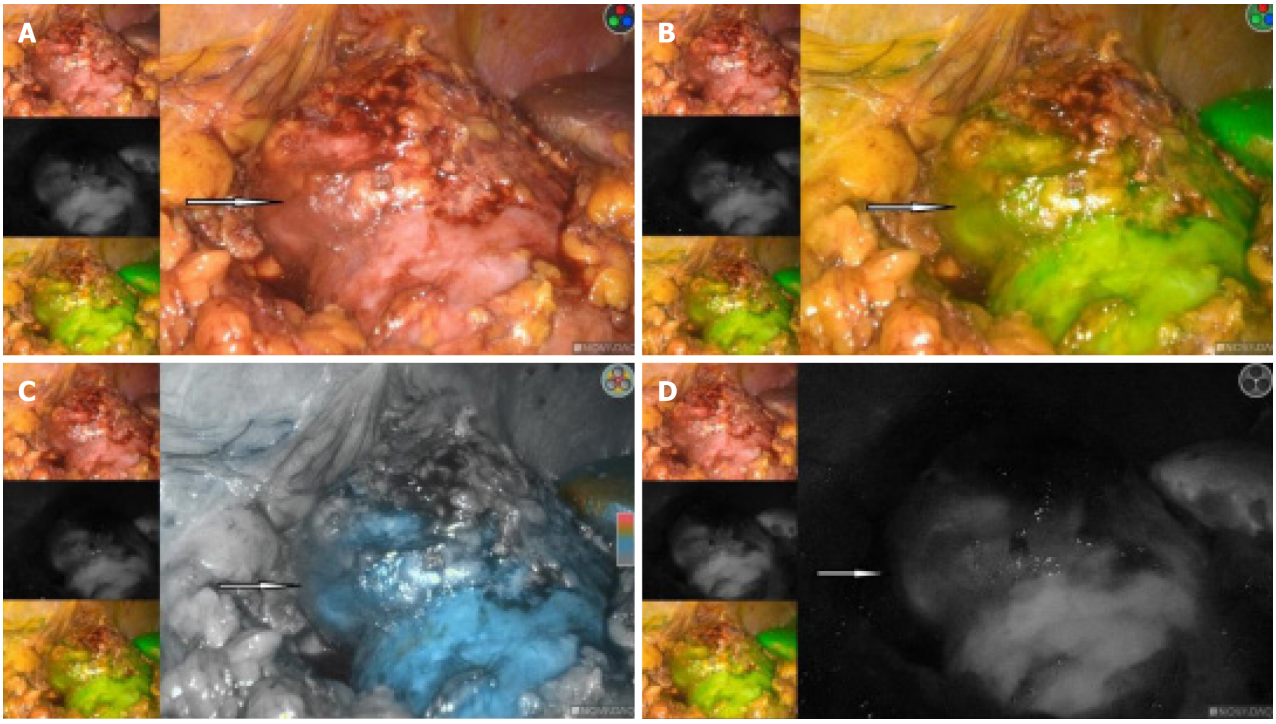


**Figure 4 In the fluorescence mode, the renal tumor had almost the same color as the surrounding normal tissue, and the border is hard to recognize.** A: In the white-light mode; B: In the green-light mode; C: In the color light mode; D: In the black and white light mode.

with ectopic kidney arterial blood supply that was enhanced on CT. The ectopic artery was emitted from the abdominal aorta, below the renal arteries. Some scholars believe that whether the renal epithelioid angiomyolipoma with independent blood supply is more aggressive behavior and malignant tendency to change, so we initially considered the mass as renal cell carcinoma, and tended to undergo radical nephrectomy.

B-ultrasound is the preferred screening and diagnostic method for renal tumors, but it is still difficult to identify some atypical cases due to its limited display of tiny blood vessels and blood flow in the lesion[11]. Intraoperative ultrasonography (US) is a commonly used technique for tumor differentiation during surgical procedures. In renal tumors, intraoperative US plays a crucial role in identifying and distinguishing various types of kidney masses, aiding surgeons in making real-time decisions and ensuring optimal treatment outcomes. During partial nephrectomy for renal tumors, intraoperative US helps in differentiating between malignant and benign lesions based on their characteristics such as size, shape, echogenicity, vascularity, and margins. But there are also some limitations to consider. One limitation is the potential challenge in accurately distinguishing between benign and malignant renal masses based solely on intraoperative US imaging characteristics. While certain features such as vascularity, shape, and margins can provide clues to the nature of the tumor, these findings may not always be definitive.

CT with contrast enhancement is the most commonly used radiologic method to diagnose RAML[12]. However, there are bleeding, cystic changes, small lesions or fat-poor in the tumor, and CT scan often cannot accurately measure the internal adipose tissue, which is easy to cause misdiagnosis[13]. Biphasic helical CT may be useful in differentiating RAML with minimal fat from RCC, with homogeneous tumor enhancement and prolonged enhancement pattern being the most valuable CT findings[14]. Some studies shown that the fat-poor RAML had a significantly higher mean

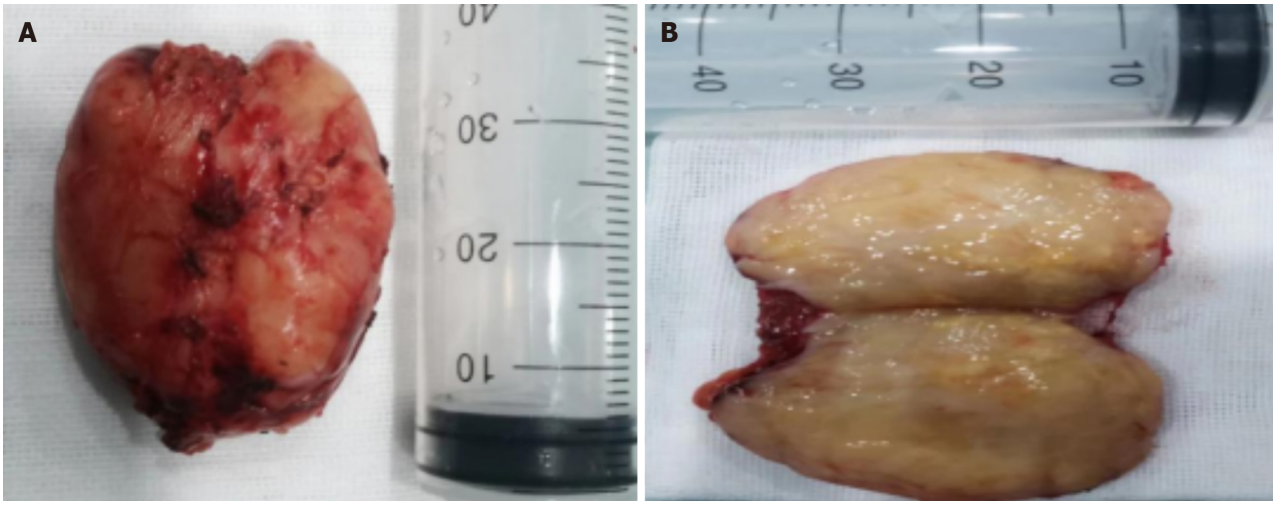


**Figure 5 Tumor images.** A and B: Picture of the right kidney tumor specimen, the tumor was successfully removed and the margin was intact.

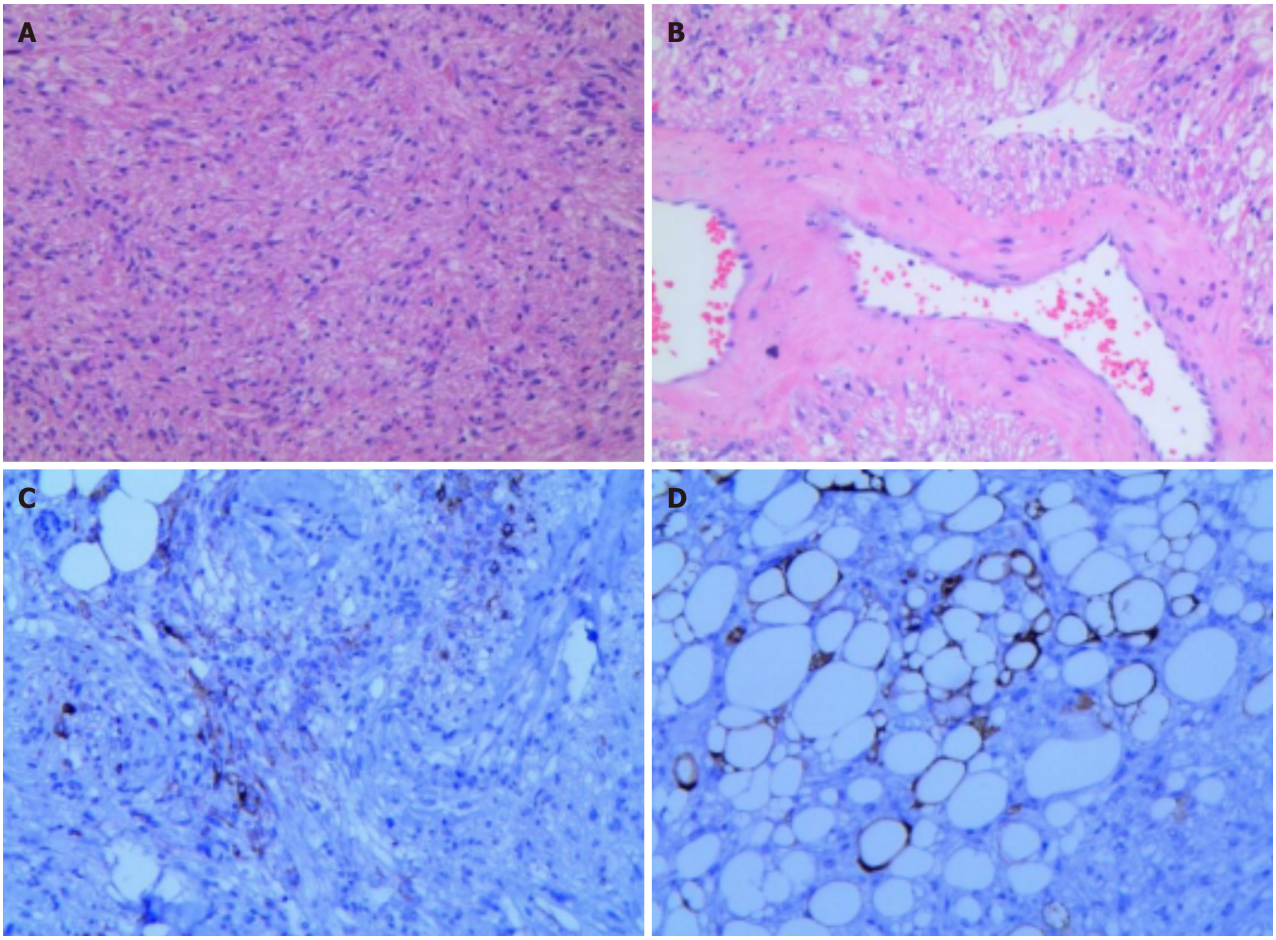


**Figure 6 Histopathological image.** A: Histopathological image showing tumor cells composed of spindle or fatty spindle cells with moderate cell density and a strip-like arrangement (H&E staining); B: Histopathological image showing significant thick-walled vessels with vitreous changes, and tumor cells seemed to distribute around the vessels; C: Immunohistochemical images showing positivity for HMB45; D: Immunohistochemical images showing S-100 positivity were suggestive of mature adipocytes.

attenuation value compared with that of renal cell carcinoma on unenhanced CT scans. In addition, significant differences were found between fat-poor RAML and RCC with regard to wash-in and enhancement ratios on contrast-enhanced CT scans[15]. Kim *et al*[16] established a diagnostic scoring system by comparing the ratio of long-to-short diameter, enhancement characteristics, tumor attenuation on unenhanced scan, tumor margin, calcification, age, and sex, to distinguish fat-poor RAML and RCC with high accuracy, sensitivity and specificity. Someone also identified RAML and RCC by intratumoral blood volume[17]. Lassel *et al*[18] combined demographic, CT enhanced characteristics on fat-poor

RAML and showed specificity > 95%, In conclusion, there is no method to confirm fat-poor RAML through a single CT image, and multiple imaging methods are needed to improve its diagnostic rate.

MRI is the examination method for distinguishing fat-poor RAML from RCC after ultrasound and CT. Currently, various MRI techniques can be used to diagnose fat-poor RAML. RCC often showed significantly hypersignal on T2 weighted imaging (T2WI), fat-poor RAML often shows low and slightly lower signal, and the signal is more uniform. Choi *et al*[19] suggested that the low signal of fat-poor RAML on T2WI correlated with the smooth muscle content in the lesion. Schieda *et al*[20] confirmed that the signal intensity (SI) of the lesion on T2-weighted MR images is important for the identification of fat-poor RAML[21]. However, Hindman *et al*[22] reported that there were no differences between minimal fat RAML and clear cell RCC for the SI index. Kang *et al*[23] noted that evaluation of apparent diffusion coefficient values can help to determine between benign and malignant lesions of renal tumors[23-25]. However, there are no characteristics MRI images of fat-poor RAML.

Studies have reported experience with percutaneous renal needle biopsy and obtained meaningful results[26], but it has not yet become a routine examination method and has concerns about the risk of causing tumor metastasis and spread. However, current technologies rely on pre- and intraoperative 3D reconstruction, which ensures safer selective arterial clamping[27]. By utilizing three-dimensional reconstruction technology, surgeons can better plan their approach, visualize the optimal clamping location, and navigate the surgical field with enhanced accuracy. This ultimately leads to a more targeted and meticulous procedure, minimizing potential complications such as excessive bleeding and post-operative renal function impairment. Although preoperative and intraoperative three-dimensional reconstruction technology offers numerous advantages in the surgical treatment of renal tumors, it also faces limitations such as challenges in professional training, time and economic costs. When using this technology, it is important to weigh the pros and cons to ensure that its advantages are maximized to enhance the safety and success rate of surgery.

In recent years, infrared fluorescence technology with indocyanine green as contrast agent has been applied in urology [28]. Some studies have reported that infrared fluorescence imaging can clearly show the size and location of renal tumors during operation, and improve the ability to deal with complex renal tumors[29]. Previous studies showed that the loss of ICG-based fluorescence only appeared in renal clear cell carcinoma[30]. Our clinical practice also presented that only renal clear cell carcinoma looked dark in the fluorescence mode. While other renal tumors looked as bright as the normal renal tissues. In this case, we used the PINPOINT fluorescent laparoscopic system intraoperatively, the tumor looked as bright as the normal renal tissues, so we performed a partial nephrectomy. The final pathology suggested a fat-poor RAML, thus avoiding nephrectomy. At the same time, we finely identified the tumor supply vessels through fluorescent navigation, thus only blocking the arteries supplying the tumor during surgery, preventing intraoperative bleeding due to malocclusion and minimizing the loss of renal function.

## CONCLUSION

Preoperative differentiation of fat-poor RAML from RCC remains a difficult imaging problem, and a percutaneous renal biopsy can be selected for the diagnosis of highly suspected fat-poor RAML. For patients who do not accept or cannot undergo percutaneous renal tumor biopsy, The use of fluorescent laparoscopy can effectively help intraoperative management, and the fluorescence pattern provided by intravenous ICG can help suggest the final diagnosis, effectively guide the surgical decision-making, and avoid preoperative imaging diagnosis leading to nephrectomy for benign renal tumors, through fluorescent navigation of tumor supply vessel precise block, minimize the loss of renal function.

## FOOTNOTES

**Author contributions:** Tang JE and Yang H contributed to the conception and design of the review; Tang JE wrote the manuscript; Fang ZH and Wang RJ validate the manuscript; Zhu PY and Yao JX contribute to the establishment of the model; All authors have read and agreed to the published version of the manuscript.

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