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**Intravenous leiomyomatosis with different surgical approaches: Three cases of report**

He J *et al*. IVL with different surgical approaches

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**Abstract**

***BACKGROUND***

Intravenous leiomyomatosis (IVL) is a rare and complicated disease, which requires surgery by a multidisciplinary team. However, the optimal surgical approach has not been determined.

***CASE SUMMARY***

Here we report three cases of IVL treated with different surgical approaches. All patients presented with the circulation symptoms. Two patients had lower extremity edema and the other had cardiopalmus. The diagnosis of IVL was confirmed based on the imagining examinations and pathological findings. All patients underwent surgical treatment and were discharged without any complication.

***CONCLUSION***

Preoperative examination is crucial for surgical planning and surgical approach is dependent on the patient’s condition and tumor involvement.

**Key words:** Intravenous leiomyomatosis; Surgery; Cardiac tumors; Case report; Minimally-invasive

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**Core tip:** Intravenous leiomyomatosis (IVL) is a rare tumor which requires surgical treatment by a multidisciplinary team. We present three cases of IVL treated with different surgical approaches, including one-stage, two-stage and minimally-invasive surgery. We describe our experience and difficulties in the surgery in order to guide the fellow surgeons in treating this rare disease.

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**INTRODUCTION**

Intravenous leiomyomatosis (IVL) is a rare and benign tumor originating from female genitalia and extending into the extrauterine vein or even the right-sided cardiac chambers[[1](#_ENREF_1)]. Approximately 300 cases[[2](#_ENREF_2)] of IVL have been reported in the English literature since it was first reported by Hirshfield in 1896[[3](#_ENREF_3)]. IVL comprised only 0.097% to 0.26%[[4](#_ENREF_4),[5](#_ENREF_5)] of all female leiomyomatosis in different institutes. The average age of IVL presentation is 45 years[[6](#_ENREF_6)]. Most patients are asymptomatic even with cardiac involvement. Surgical treatment is recommended for all patients. Due to massive extension of the tumor, multidisciplinary cooperation is required in the surgery. To date, there is still no universal consensus regarding the optimal surgical approach[[1](#_ENREF_1)]. Here we present three cases of IVL and outline our experience in the surgical treatment and outcome.

This study was approved by the Ethics Committee of Guangdong General Hospital and written consent was obtained from the patients.

**CASE PRESENTATION**

***Chief complaints***

**Case 1:** A 46 year-old nulliparous Chinese woman presented with chest distress and lower extremity edema for 1 mo.

**Case 2:** A 46-year-old multiparous Chinese women presented with cardiopalmus for 2 mo.

**Case 3:** A 54-year-old multiparous Chinese women presented with lower extremity edema for 1 year.

***History of present illness***

**Case 1:** The patient complained of chest distress and short of breath after activity 2 mo ago. She had no chest pain, cough and paroxysmal nocturnal dyspnea. The symptoms aggravated and combined with lower extremity edema. She came to our hospital for future treatment. Her appetite, sleep, voiding and stool were normal. No obvious weight and physical strength change.

**Case 2:** The patient presented with cardiopalmus for 2 mo. The symptom can relieved after few minutes rest. She had no chest distress, chest pain. The echocardiography in local hospital detected multiple mass in the right heart chamber. She came to our hospital for future treatment. Her appetite, sleep, voiding and stool were normal. No obvious weight and physical strength change.

**Case 3:** The patient complained of lower extremity edema 1 year ago. The symptom was associated with menstruation and aggravated in the last 2 mo. Therefore, she came to our hospital for help. She also had abdominal distention but no abdominal pain or nausea. Her appetite, sleep, voiding and stool were normal. No obvious weight and physical strength change.

***History of past illness***

**Case 1:** The patient denies a history of hypertension, diabetes and hepatitis. She denies any surgery or injury.

**Case 2:** The patient had a history of hysterectomy for uterine leiomyoma. She denies hypertension, diabetes and valvar heart disease history. Deny the injuries.

**Case 3:** The patient had a history of hypertension which had not been properly monitored or treated. She denies other diseases, surgery and injury.

***Personal and family history***

**Case 1:** She does not smoke and drink. No exposure history to toxic substances and infected water. Her menstruation was normal.

**Case 2:** She never smoke and drink. No exposure to toxic substances and infected water. She denies family history of tumor. Her menstruation was stop.

**Case 3:** She has no chance to contact with poison. She does not smoke or drink. No infective and hereditary diseases. She denies the family history of such disease.

***Physical examination upon admission***

**Case 1:** Temperature (T): 36.5℃; Pulse (P): 75 bpm; Blood pressure (Bp): 128/82 mmHg; Weight (W): 47.5 kg. No heart murmur was noted during auscultation. Breaths sounded clear, and no rale was noted. No peripheral vessel sign was noted. No lower extremity edema was detected.

**Case 2:** T: 36.5℃; P: 85 bpm; Bp: 110/70 mmHg; W: 57 kg. No heart murmur was noted during auscultation. Breaths sounded clear, and no rale was noted. No peripheral vessel sign was noted.

**Case 3:** T: 37.1℃; P: 98 bpm; Bp: 142/88 mmHg; W: 52 kg. No heart murmur was noted during auscultation. Breaths sounded clear, and no rale was noted. No peripheral vessel sign was noted. No lower extremity edema was detected.

***Laboratory examinations***

**Case 1:** Blood test showed ALT 58.85 U/L, BNP 140 pg/mL, E2 26 pg/mL, HE4 78.46 pmol/L and β-HCG 3.28 mIU/mL. Other laboratory results were within normal limits.

**Case 2:** Blood test showed TBIL 20.9 μmol/L and BNP 670.1 pg/mL. Other laboratory results were within normal limits.

**Case 3:** Blood test showed WBC 11.34 × 109/L, Hb 97.6 g/L, E2 195 pg/mL, β-HCG 3.28 mIU/mL and BNP 672.1 pg/mL. Other laboratory results were within normal limits.

***Imaging examinations***

**Case 1:** Computer tomography (CT) showed a mass (Figure 1A) in the pelvis encroaching the uterus and involving the right internal iliac vein, the inferior cava vein (ICV) and the right atrium (RA). Echocardiography detected large immobile echogenic mass attaching to the atrial septum (Figure 1B).

**Case 2:** CT showed soft tissue within left uterine adnexa-left ovarian vein-left renal vein-ICV-RA-right ventricle (RV) and no involvement of portal vein and its branches was detected (Figure 2A). Echocardiography revealed massive continuous mass extending from IVC through the RA and crossing into the RV (Figure 2B). Her vital sign was stable and no specific change was found. A biopsy was performing to confirm the diagnosis of IVL.

**Case 3:** CT revealed multiple masses located in the right lower quadrant and pelvis merging together with diameter of 13 cm. The tumor reached down to the right pelvic floor, surrounded the right ureter and extended to the RA through the right ovarian vein, right common iliac vein and ICV (Figure 3A). Echocardiography showed cord-like mass crossing into the RV through tricuspid valve (Figure 3B).

**MULTIDISCIPLINARY EXPERT CONSULTATION**

***Case 1***

**Guan-Di Chen, MD, PhD, Professor, Department of Gynecology:** The patient appeared the main symptom of vein occlusion. Imagining findings indicated IVL which required surgical treatment. For women above 40 years old, we recommended hysterectomy and bilateral salpingo-oophorectomy.

**San-Ming Wang, MD, PhD, Professor, Department of Vascular Surgery:** There were multiple lesion from the right iliac vein to the RA. According to the imaging findings, IVL is the most likely diagnosis. As the tumor invade the RA, surgical treatment is necessary.

**Cong Lu, MD, PhD, Chief Doctor, Professor, Department of Cardiovascular Surgery:**I concurred with the above opinions. IVL is the most likely diagnosis. But the possibility of malignant tumor still could not be ruled out. We recommended gynecologic surgery first and when the IVL was confirmed after the pathological exam, a second operation would be scheduled.

***Case 2***

**Mu-Biao Liu, MD, PhD, Professor, Chief Doctor, Department of Gynecology:** According to the patient history and preoperative exam, the diagnosis of IVL was confirmed. Bilateral salpingo-oophorectomy was recommended for this patient.

**Gang Zhao, MD, PhD, Professor, Chief Doctor, Department of Vascular Surgery:** The diagnosis of IVL was confirmed and surgical indication was clear. No attachment was detected and the invasion of the tumor was clear. There was a good chance to remove the tumor completely at once. In our experience, extraction from both the RA and the IVC might provide better chance of complete resection and bleeding control.

**Jin-Song Huang, MD, PhD, Chief Doctor, Professor, Department of Cardiovascular Surgery:** I agreed with the above views. The patient general health condition was suitable for one-stage operation.

***Case 3***

**Gang Zhao, MD, PhD, Professor, Chief Doctor, Department of Vascular Surgery:** According to the imaging findings, the most likely diagnosis is IVL. CT revealed the tumor in the pelvic is huge and the operation to completely resect the tumor could be time-consuming and the chance for massive bleeding was very big. Therefore, two-stage operation was recommended.

**Huan-Lei Huang, MD, PhD, Chief Doctor, Professor, Department of Cardiovascular Surgery:**The mass in the RA was large but mobile. No attachment to the RA was detected. There was a good chance to completely remove the mass.

**Hai-Yan Ye, MD, PhD, Professor, Department of Gynecology:** I agreed with the above opinions. The mass in the pelvic was very big with rich blood supply. It would be very difficult to completely resect the tumor. A two-stage operation might be beneficial.

**FINAL DIAGNOSIS**

The diagnosis of IVL in these three patients was confirmed based on the imagining examinations and pathological exam.

**TREATMENT**

***Case 1***

This patient was scheduled for two-stage operation. Due to the potentially malignant nature of the tumor, laparotomy was performed first. The uterus was slightly enlarged with uneven surface and a nodular mass arose from it. The right uterine vein engorged. Total hysterectomy and bilateral salpingo-oophorectomy was performed. The operation went on smoothly. Histologic examination of the tumor confirmed the diagnosis of leiomyoma. After 14-d of routine treatment, the patient recovered well and underwent a second-stage operation. The heart was exposed through a median sternotomy. Cardiopulmonary bypass (CPB) was instituted by ascending aorta and superior vena cava cannula and systemic cooling was commenced. Laparotomy was performed at the same time. The right common iliac vein and ICV were exposed after meticulous dissection. The right and left renal veins were controlled with vascular clamps. Myocardial protection was achieved by cold blood cardioplegia. Then, venotomy was made on ICV at the level of renal veins. In the meantime, right atriotomy was performed. The tumor exposed on the ICV incision was transected. The upper part of the tumor was extracted from the RA and the rest of it was extracted from ICV (Figure 1D). The whole extraction process was very smooth. The incision on the RA and ICV were closed separately. The operation time of these two surgeries were 165 and 190 min respectively, and the blood loss was 200 and 500 mL respectively.

Postoperative pathological examination showed mass smooth muscle fibers proliferation with interstitial fibers edema and a few lymphocytes infiltration. No cellular atypia was observed (Figure 1C). The rest of the surgery was uneventful and postoperative treatment was uncomplicated.

***Case 2***

The patient was scheduled for one-stage operation. After general anesthesia, simultaneous sternotomy and laparotomy was performing. A mass was seen in the right pelvis with diameter of 4 cm surrounding the left uterine adnexa. The left ovarian vein enlarged with diameter of 2 cm containing cord-like mass which extended to the IVC through the left renal vein. Total hysterectomy and bilateral salpingo-oophorectomy was performed and the distal end of the left uterine vein was ligated. The vein containing the mass was isolated to the level of left renal vein. The right posterior peritoneum was opened and the ICV was fully exposed. CPB was institute by ascending aorta and superior vena cava cannula and systemic cooling was commenced. Renal veins were controlled with vascular clamps and incisions were made on the RA and IVC. Then, the mass was transected through the ICV incision. The lower part of the mass was extracted successfully. When we tried to extract the upper part of the mass, there was obstruction in the RV and the ICV. Serve bleeding occurred in the ICV and the blood pressure dropped dramatically. Systemic cooling to a deeper temperature was commenced. After exploration, we found adhesion to the RV and part of the tumor extending into the portal vein causing obstruction. We cut off the adhesion along with some of tendon from RV and sharply separated the tumor with portal vein leaving part of the mass (Figure 2D). The incisions were closed quickly and cannula on the IVC was placed to maintain the flow of CPB. The blood pressure went up gradually. No tricuspid regurgitation was detected by the transesophageal echocardiography (TEE) during surgery. The rest of the surgery was uneventful. The whole operation took 600 min and the blood loss was 4000 mL.

Postoperative pathological examination showed concentration of spindle cell with mass hyaline degeneration. Immunohistochemistry revealed SMA (+++), Caldesmon (+++), ER (+++) and PR (+++). (Figure 2C)

***Case 3***

The patient was scheduled for two-stage operation. During the first surgery, laparotomy was performed. A hug mass was seem in the pelvis reaching the sacrum on the top and attaching to the pelvic floor. The uterus, right oviduct and right ovary were invaded by the tumor. The bladder and the right ureter were attached to the tumor. The tumor in the pelvis was excised completely meanwhile both adnexa uteri were removed. The whole operation took almost 7 h and there were approximately 1200 mL blood lost. After 2 wk recovery, the patient underwent the second operation. Laparotomy was performed. The abdominal adhesion was divided and the ICV was exposed. The CPB was instituted by femoral artery, femoral vein and jugular vein cannula. Two small incisions were made on the right chest at the midclavicular line and maxillary line separately. The CPB was commenced on normal temperature. Atriotomy and venotomy were performed at the same time. The tumor was transected and extracted from the atrium and the ICV. During extracting the tumor on the bottom, there were some adhesions to the vein wall. We clamped the proximal end of the tumor and separated the tumor following the direction of the vein. We took out the tumor along with part of the vein intima causing tear on the ICV incision (Figure 3D). The incisions were closed with 5-0 prolene and the CPB was disconnected. No residual tumor was detected by the TEE in the RA. The rest of the operation was uneventful. The time of these two operations were 395 and 265 min and the blood loss was 1200 and 2000 mL respectively.

Postoperative pathological examination showed concentration of smooth muscle cell with local hyaline degeneration. Immunohistochemistry revealed P16 (-), P53 (-), Ki67 (10 %+), Caldesmon (+++) and CD10 (±). (Figure 3C)

**OUTCOME AND FOLLOW-UP**

***Case 1***

Postoperative treatment was uneventful. She was discharged 5 d after the second surgery. Follow-up CT did not detect any abnormality. She remained asymptomatic since then.

***Case 2***

Postoperative vasoactive agent and antibiotics were routinely used. She was discharged 21 d after the surgery without any complication. The follow-up CT in 3 mo after discharge detected filling in retrohepatic segment of ICV.

***Case 3***

Postoperative treatment was uneventful. She was discharged 11 d after the second surgery. Follow-up CT did not detect any residual tumor.

**DISCUSSION**

IVL is a rare and potentially fatal disease, with a juxtaposition of benign histopathologic features and quasi-malignant clinical behavior. To date, approximately 300 cases of IVL have been reported[[2](#_ENREF_2)]. Clinical symptoms are nonspecific and clinical diagnosis mostly relied on radiology and pathology examinations. Surgery is still the gold standard for treatment of IVL. Due to the extensive nature of the tumor and its involvement in multiple systems[[7](#_ENREF_7)], multidisciplinary cooperation is required for its treatment. However, there are still no guidelines regarding the surgical approach for IVL. In this study, we reported 3 cases of IVL treated with different surgical approaches. We intend to share our experience and provide valuable guidance for fellow surgeons in treating this rare disease.

Surgery is currently the only effective treatment for IVL and complete tumor excision is a valid treatment for the prevention of recurrence[[8](#_ENREF_8)]. The surgical difficulties in IVL are mainly associated with the adhesion between the tumor and the venous wall rather than the length of the tumor. Careful preoperative examination for evaluation of the adhesion, especially in the retrohepatic segment of the IVC and the RV is essential, and provides valuable information for surgical planning. Both one-stage and two-stage operation are viable treatment options depending on the patient’s general condition and invasion of the tumor. Recently, the medical community prefers an initial thoracic approach, which allows safe resection and reduces the risk of tumor dislodgement and pulmonary embolism. However, we have a different point of view. We believe that a one-stage operation using an abdominal approach could result in better control of the tumor and a better chance of complete resection.

In Case 1, preoperative examination did not rule out the possibility of malignant tumor. Therefore, we chose a two-stage operation. However, this is certainly not a criterion for two-stage operation. The operation was very successful and the tumor was completely resected.

In Case 2, we performed a one-stage operation as extensive adhesion was not detected on preoperative examination. However, when we tried to extract the tumor, occlusion in both the IVC and the RV was observed. Subsequent massive hemorrhage occurred in the IVC and the patient’s blood pressure dropped dramatically. We immediately commenced systemic cooling to provide better brain protection. The adhesion was then removed leaving part of the tumor in the IVC, the incisions were closed and another cannula was placed in the IVC to maintain the circulation. The remainder of the procedure was uneventful. In cases where the mass is difficult to extract, severe bleeding may occur at the IVC incision or the RA incision and the circulation is difficult to maintain. Therefore, proactive placement of a cannula in the femoral vein may be beneficial. The follow-up CT also confirmed adhesion in the IVC, especially in the retrohepatic segment.

In Case 3, the tumor in the pelvic was huge and invaded the urinary system, which might lead to massive blood loss and trauma. Thus, we chose a two-stage operation. In the second operation, we used a minimally-invasive approach in the chest as there were fewer adhesions in the right cardio-chambers. The time of the open heart procedure was estimated to be short. We employed normothermic CPB with no cardiac arrest, which minimized trauma and disturbance in the circulation. This is the first time that minimally-invasive surgery has been performed in IVL. However, when we tried to extract the remainder of the tumor from the IVC incision, the tumor could not be removed. We separated the adhesion and removed the tumor along with part of the IVC intima, subsequently causing a tear in the IVC. There are two theories regarding the origin of IVL[[2](#_ENREF_2)]. One is that the IVL arises from the myometrium, invades the vein and grows along with it. The other theory is that the IVL is a leiomyoma arising directly from the venous wall and is difficult to extract. The latter theory may explain the situation we encountered. Nonetheless, there is another explanation as we believe that ligation or sutures from the previous operation may have caused the obstruction. In such cases, we recommend proactively introducing two sutures on the venous wall to prevent future tearing and this will result in easier incision closure. It is also worth mentioning that the venous wall is very delicate and easily damaged. Extra care should be taken when trying to control the vein with a clamp. Pathological examination of tissue from all three cases confirmed the diagnosis of IVL and no other specific characteristics were found.

Mini-invasive surgery is a trend in cardiovascular surgery. It can save patient from sternotomy and reduce the surgical trauma. But there is still limitation in its application in cardiovascular surgery and this is the first attend in IVL. Before the surgery, the mobility of the mass was assessed by the echocardiography and no attachment was found (especially in the hepatic vein). We made three small incisions in the chest and started the CPB at normal temperature without cardiac arrest. We transected the tumor at the level of IVL incision in the abdomen, leaving the upper part of the tumor not too long for extraction. The extraction process in the chest went smoothly and the CPB time was only 52 min, which is relatively shorter than other report[[9](#_ENREF_9)]. During the surgery, we used TEE to make sure that there was no residual tumor in the RA. The mini-invasive approach in the chest can reduce the surgical trauma, greatly preserve the cardiac function and save some operation time.

Multidisciplinary approach, whether in one-stage or two-stage fashion, is considered to be safer and becomes a common practice[[10](#_ENREF_10)]. The primary indication for choosing the stage operation, in our opinion, is invasion of the tumor in the pelvis and the general condition of the patient. Since the whole operation for IVL can be time-consuming, the surgical trauma can be very severe. Especially when the left ovarian vein is involved, we need to open the posterior peritoneum in both sides, which may resulted in extensive diffuse blood oozing after the CPB or even deep hypothermic circulatory arrest. In such case, we recommend two-stage operation.

As for the pros and cons of the one-stage and two-stage operation, one-stage operation can reduce the risk of a second anesthesia[[11](#_ENREF_11)], the psychological burden of the patient and the risk of tumor embolism. But it might increase the operation time and the trauma at one time. In the meanwhile, two-stage operations can relatively manager the surgical trauma. But the abdominal adhesion from the first operation might increase the difficulty. In our mind, we prefer the one-stage operation if the criterion is met.

In this series, the mean operation time is 571.7 min, which is consistent with other report[[9](#_ENREF_9)]. The blood loss in the last two cases is considerably big due to the adhesion to vein wall and subsequently mass hemorrhage. It is worth mentioning that the CPB time in case 3 is relatively shorter than other report, which might indicate the effectiveness of mini-invasive approach in IVL.

The pathological exam and the immunohistochemistry revealed the same characters as reported by Tang[[2](#_ENREF_2)], classical leiomyomata with uniform spindle cells arranged in intersecting fascicles. Only a few mitotic figures are seen without cytologic atypia. The tumor is positively immunolabelled with SMA and Caldesmon, and negative with P16 and P53. Staining for ER and PR is strongly positive.

Hormonal therapy for IVL patients is still controversial. The most common practice of hormonal therapies is used as a neoadjuvant for the patients with incomplete resection or who do not have surgery. The commonly used agents are GnRH agonists, tamoxifen and medroxyprogesterone[[12](#_ENREF_12)]. The clinical outcomes are mixed. Four studies reported minimal or no response to the therapy[[13-16](#_ENREF_13)], while some reported meaningful response[[17-19](#_ENREF_17)]. Even in a comprehensive analysis by Li[[10](#_ENREF_10)], hormonal therapy is considered invalid in preventing recurrence. In this series, there was tumor residual detected by follow-up CT in case 2. Since the growth of the tumor is very slow, we decided to keep her in close follow-up without hormonal therapy. Recurrence of IVL is pretty rare once the tumor was completely resected. There are patients remaining free of disease 11 years after the surgery[[20](#_ENREF_20)]. Only one case of suspected pelvic recurrence has ever been reported[[21](#_ENREF_21)].

In conclusion, we report three cases of IVL treated with different surgical approaches and highlight the outcomes of these surgical techniques. Surgery is the only effective option for IVL. With regard to the specific surgical approach for IVL, whether a one-stage or two-stage, minimally-invasive or traditional approach is used will depend on the patient’s condition and tumor involvement. Preoperative examination is crucial for surgical planning.

**CONCLUSION**

(1) Preoperative examination is crucial. It can tell us the involvement and adhesion of the tumor, which is important in surgical planning; (2) The invasion of the tumor in the pelvic is an important factor to factor to decide one-stage or two-stage operation; (3) Extraction of the tumor from the RA and IVC simultaneously might be beneficial, as it can provide better bleeding control and complete excision; and (4) Mini-invasive approach in the chest might be a safe and effective procedure.

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**Figure 1** **Representative images of Case 1**. A: Computer tomography scan shows a continuous mass arising from the pelvis through the inferior cava vein; B: Echocardiography shows the mass extending into the right ventricle from the right atrium; C: Pathological examination of the tumor showed mass smooth muscle fibers proliferation with interstitial fibers edema and a few lymphocytes infiltration; D: The resected tumor during the surgery.

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**Figure 2** **Representative images of Case 2**. A: Computer tomography scan shows a continuous mass arising from the left side of the pelvis through the left renal vein and the inferior cava vein; B: Echocardiography shows the mass extending into the right ventricle from the right atrium; C: Pathological examination of the tumor showed concentration of spindle cell with mass hyaline degeneration; D: The resected tumor (displayed from the top to the bottom).



**Figure 3** **Representative images of Case 3**. A: Computer tomography scan shows a continuous mass arising from the pelvis through the inferior cava vein; B: Echocardiography shows the mass in the right cardiac chamber; C: Pathological examination of the tumor showed concentration of smooth muscle cell with local hyaline degeneration; D: The resected tumor (displayed from the top to the bottom).