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CASE REPORT

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Single-stage resection of uterine fibroids and intravascular leiomyomatosis: a case report

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Abstract

Background Uterine Fibroids (UFs) are common benign tumors in the female reproductive tract, but their progression to intravascular leiomyomatosis (IVL) is rare. Presently, there are few reports on single-stage resection of UFs and IVL.

Case presentation A 47-year-old woman, G2P2, had been diagnosed multiple UFs four years ago and now developed heart failure. Imaging examinations revealed that UFs had invaded the right iliac vein and extended into the right atrium through the inferior vena cava. Through multidisciplinary collaboration and a single-stage resection, the patient has survived for over 24 months post-surgery, and her heart function has significantly improved compared to preoperative levels, with no recurrence of UFs observed.

Conclusions Single-stage resection of IVL and UF is feasible and advantageous for this case, and selecting the appropriate surgical approach is crucial.

Keywords Uterine fibroids, Intravascular leiomyomatosis, Heart failure

Background

Uterine Fibroids (UFs) are the most common benign tumors in the female reproductive system. In some cases, UF tissue can infiltrate veins, and potentially extend into the heart and pulmonary blood vessels. When a mass is found in the inferior vena cava or heart, UF should be considered as a possible source. UFs that grow in veins are called intravascular leiomyomatosis (IVL). IVL is a rare benign tumor, and the mechanism of occurrence and development of IVL is currently unclear. Due to the impact of IVL on hemodynamics and the risk of sudden cardiac function deterioration, timely selection of appropriate surgical plans and timing is crucial. Herein, we successfully treated a UF case with IVL through a single-stage resection. Figure 1 shows the diagnosis and management process of the patient.

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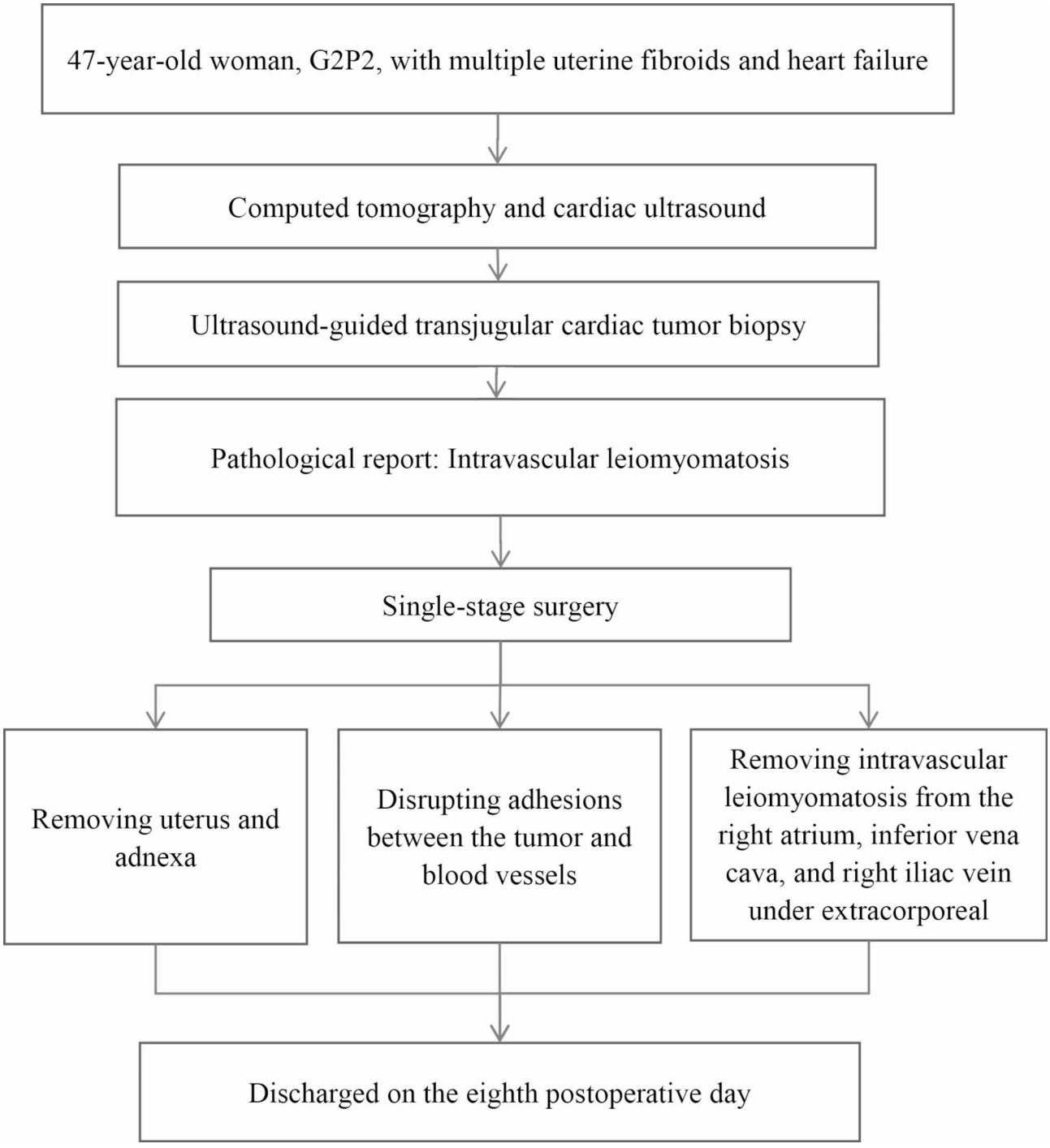

Fig. 1 Flowchart of diagnostic and management

Case presentation

A 47-year-old woman, gravida 2 para 2 (G2P2), was diagnosed with multiple Uterine Fibroids (UFs) in a routine physical examination four years before current admission. Recently, the largest fibroid measured approximately 10×12 mm by an ultrasound examination. Two months before admission, she visited the obstetrics and gynecology department of the district general hospital in the urban setting due to abnormal vaginal bleeding. A transvaginal ultrasound showed multiple UFs, including intramural (45×50×43 mm), submucosal (30×22×16 mm), and subserosal (114×54×48 mm and 90×85×28 mm) growths. The shape and position of the intrauterine device placed two decades ago were normal. Computed tomography (CT) and cardiac ultrasound revealed filling defects in the right atrium (RA) (Fig. 2A,

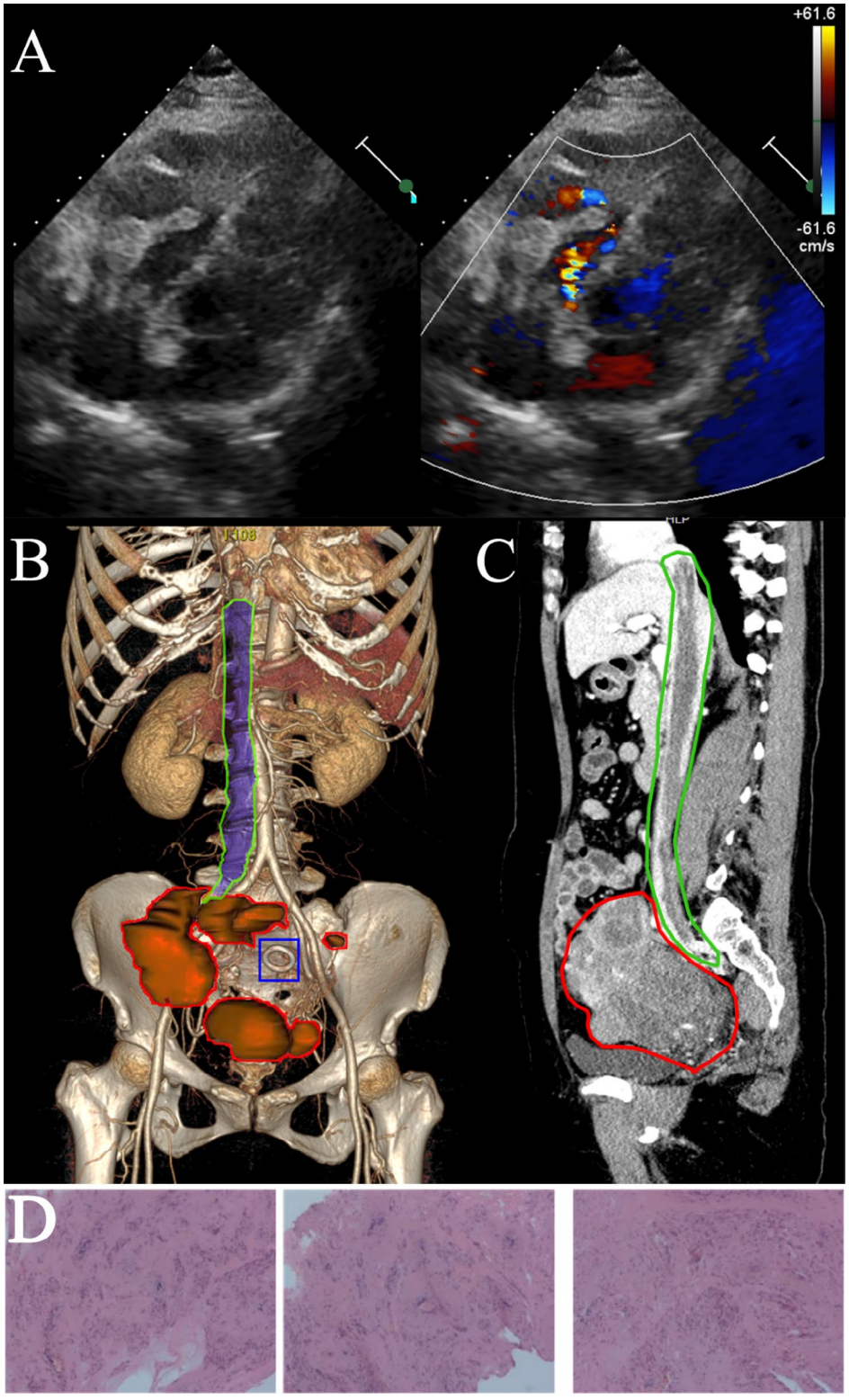

Fig. 2 (A) Preoperative cardiac ultrasound of the patient; (B) Preoperative CT three-dimensional reconstruction of the patient. The block shaped object marked with a red line represents multiple Uterine Fibroids, the blue box indicates an intrauterine device (IUD) inserted by the patient 20 years ago, and the green border represents a tumor that extends to the right iliac vein and inferior vena cava; **(C) Preoperative CT angiography of the patient.** The block shaped object marked with a red line represents multiple Uterine Fibroids, and the green border represents a tumor that extends to the right iliac vein and inferior vena cava; **(D) Preoperative tumor pathology**

B, C). Subsequently, we performed an ultrasound-guided transjugular cardiac tumor biopsy. Postoperative pathology confirmed that the mass in the right atrium and vein was an intravascular leiomyomatosis (IVL) (Fig. 2D). Further investigation of the patient's medical history revealed symptoms of chest tightness and shortness of breath after moderate activity, indicating that the tumor had migrated to the heart, causing cardiac dysfunction. Moreover, the patient's tumors in the RA could freely float, which could lead to blockage of the atrioventricular cavity and valves, circulatory embolism and sudden cardiac death. Given the potential serious consequences and complexity of the condition, we reviewed existing literature and quickly assembled a multidisciplinary team, including gynecology, cardiac surgery, vascular surgery, anesthesia, and intensive care unit [1]. Before determining the final surgical method, pathological and imaging results had already clarified the origin and characteristics of the cardiac tumors. After thorough evaluation, the team decided to perform a single-stage resection on the patient. Gynecologist consulted relevant literature and pointed out that the recurrence rate of UFs can reach 40%, and if the tumor recurs, it is likely to develop into IVL again [2, 3]. Therefore, complete resection of the uterus and adnexa is necessary. According to CTs and ultrasounds, physicians in cardiac surgery and intensive care unit believe that no other organs were involved. IVL is currently the first consideration for heart failure in the patient, due to the absence of acute chest pain and other abnormalities in blood indicators, pulmonary embolism, bronchitis, coronary atherosclerotic heart disease and other diseases can almost be completely ruled out [4]. The single-stage resection will sequentially involve hysterectomy, vascular wall tumor release, and IVL removal under extracorporeal circulation.

On the day of surgery, the obstetrician and gynecologist began by making a midline incision in the patient's lower abdomen. They then incised the peritoneum, and released the ligaments and adhesions between the uterus and surrounding pelvic tissue, thereby restoring the normal anatomical structure of the uterus and bilateral appendages. Due to multiple tumors and significant adhesion to the uterine tissue, the uterus and adnexa were completely removed. Gross pathology examination revealed four tumors, the largest measuring about 11 cm in diameter. The intrauterine device remained intact and embedded in the uterus. UFs had invaded the right iliac vein and inferior vena cava (IVC). Immediately, the vascular surgeon then proceeded with the second step of surgery. They separated and cut open the vein walls of the right internal iliac vein, right common iliac vein, and IVC, aimed at disrupting adhesions between the tumor and blood vessels. Finally, the cardiac surgeon performed the third step of surgery. After sternotomy and suspension, an ultrasound guided artery catheter was inserted into the left common iliac vein through the left femoral vein. Extracorporeal circulation was established after systemic heparinization. Upon incising the beating heart's RA, it was found that the tumor capsule was intact without obvious adhesion. The tumor extended into the heart and was extracted, along with the free tumor in the IVC and right iliac vein, measuring approximately 18 cm in length (Fig. 3A). Finally, the right internal iliac vein was then ligated. Intraoperative frozen pathology confirmed the lesion as a uterine leiomyoma (Fig. 3B). After examining the pelvic cavity, RA, and IVC, protamine was administered to neutralize heparin and stop the bleeding. The chest and abdominal cavities were then closed. The patient lost 700 ml of blood during the operation.

Following the successful completion of surgery, the patient was transferred to the intensive care unit for postoperative care, where she received medication. Including dopamine and norepinephrine to maintain blood pressure stability, corticosteroids reduce myocardial edema, furosemide for diuresis, cefoperazone sulbactam sodium for anti infection, etc. Within the first 7 days after surgery, we administered 2 g of cefoperazone sulbactam sodium every 8 h to prevent infection. For anticoagulation, 1500 U of heparin sodium was added to 250 ml of physiological saline and given as an intravenous drip once a day for 3 days. Figure 4 shows the trend of changes in the patient's coagulation function and white blood cell levels. On the third postoperative day, she was extubated and transferred to a regular ward for further treatment. She was discharged on the eighth postoperative day. Three months later, a follow-up examination showed no signs of tumor recurrence or vascular embolism on cardiac and abdominal ultrasounds. The patient has now survived for more than 24 months without any symptoms of heart failure.

Discussion and conclusions

Uterine Fibroids (UFs) are common benign tumors in the female reproductive tract. However, occurrences of uterine leiomyomas with intracardiac growth are exceedingly rare. Our literature search identified fewer than 400 reported cases since the 1900s. Previous reports indicate that over 50% of these cases opted for two-stage resection. This approach involves addressing chest and abdominal tumors in the first surgery and subsequently treating pelvic tumors in a second stage [5]. Recent small-sample studies suggest no significant difference in tumor recurrence rates between patients undergoing single-stage and two-stage resections. We have collected case reports on the treatment of intravascular leiomyomatosis (IVL) in the past five years and summarized the treatment methods and experiences from these articles (Supplementary Table 1). Both methods show good therapeutic effects

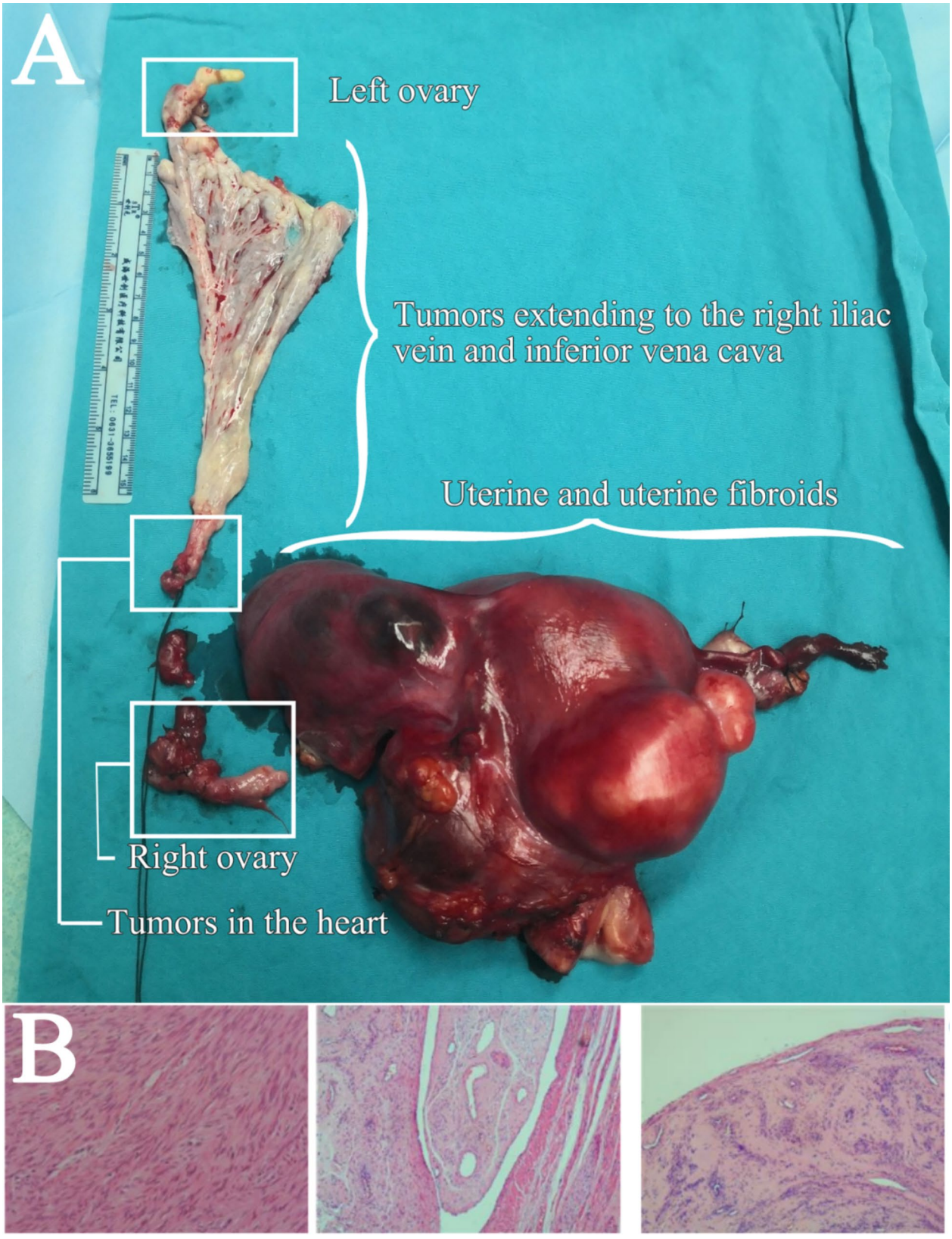

Fig. 3 (**A**) Tumor tissue removed during surgery. (**B**) Intraoperative frozen pathology

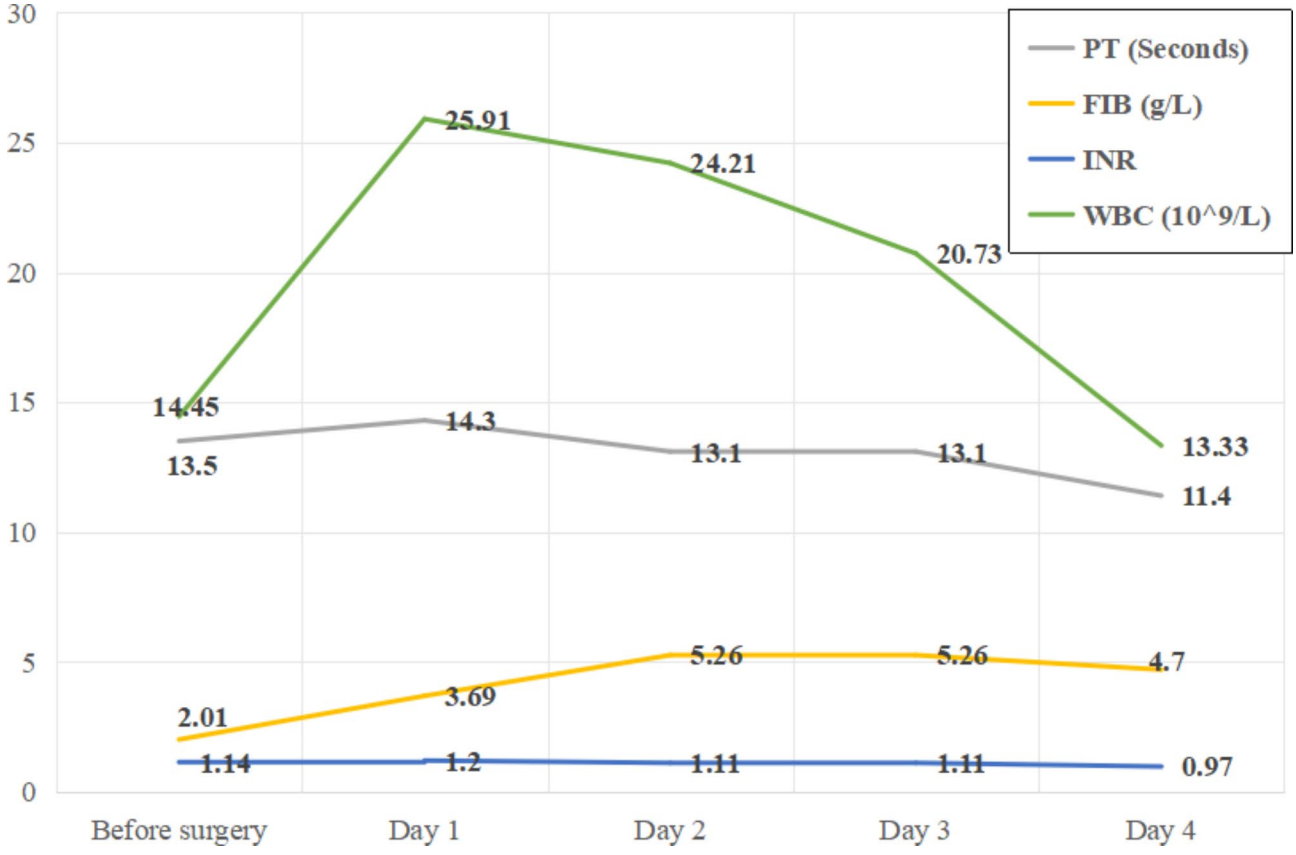

Fig. 4 Coagulation function and white blood cell levels of preoperative and postoperative patients

[6, 7]. It is worth noting that in the earliest case reports, surgeons emphasized that complete removal of the UF and IVL was key to successful treatment [8]. Single-stage resection may have more advantages than two-stage resection, which often results in more intraoperative and postoperative bleeding, as well as greater surgical trauma [6, 9]. In addition, for two-stage resection, the UF/IVL removed in the early stage may recur, and early resection may cause varying degrees of adhesion in the abdominal cavity [10, 11]. Therefore, it is crucial to fully evaluate the indications for single-stage resection in patients before proceeding. In this case, although pathologically confirmed as a benign tumor, its development exhibited characteristics indicative of malignancy, with a risk of detachment of the freely floating right atrial leiomyoma. Considering the patient's acceptable general condition and preserved heart function, we promptly decided to proceed with single-stage resection and successfully completed the resection.

This patient was previously diagnosed with UFs, but did not receive treatment. The UFs invaded blood vessels and affected blood return to the inferior vena cava, increasing the preload on the right heart. Long term insufficient cardiac output led to heart failure symptoms. For patients with uterine leiomyoma and intracardiac growth, a comprehensive preoperative evaluation should be conducted to assess whether they can withstand simultaneous thoracic, abdominal, and pelvic surgery. Doctors should also evaluate the risk of tumor detachment and embolism within the circulatory system to ensure a favorable postoperative prognosis. In cases of UFs coexisting with cardiac insufficiency, vigilance for tumor invasion into the circulatory system is necessary. This requires thorough cardiac and vascular ultrasound examinations, as well as chest, abdominal, and pelvic CT angiography to clarify tumor growth patterns and develop treatment strategies promptly. It should be noted that the recurrence rate of IVLs within 5 years after surgery is as high as 30%, and regular follow-up is important to improve patient prognosis [12].

The successful outcome of this case highlights the importance of regular gynecological examinations for women with an intrauterine device, particularly those with UFs and concurrent heart failure. This emphasizes the need for vigilance regarding fibroid growth within cardiac cavities and veins.

Abbreviations

CT Computed tomography
IVC Inferior vena cava
IVL Intravascular leiomyomatosis

RA Right atrium
UF Uterine Fibroids

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Supplementary Information

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Supplementary Material 1

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Author contributions
JX.L and WQ.Z wrote the manuscript. CH.L, JL.W and CL.W reviewed and interpreted imaging findings. J.Q and XC.Z drew the figures. LL.H, BS.Z, HF.Z and T.Z revised the manuscript and figures.

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Data availability
The datasets used and/or analysed during the current study available from the corresponding author on reasonable request. All data are obtained through retrospective analysis of medical records and do not require a Clinical Trial Number.

Declarations

Ethics approval and consent to participate
The scheme of this report was approved by the Ethics Review Committee of the First Affiliated Hospital of Guangxi Medical University. Written informed consent was obtained from the patient.

Competing interests
The authors declare no competing interests.

Consent to publish
Written informed consent was obtained from the participant for publication of identifying information/images in an online open-access publication.

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