Retroperitoneal lymphoma with double inferior vena cava shown using a 3D visualization model: A case report and literature review

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Abstract. Cases of a retroperitoneal tumor with double inferior vena cava (IVC) are rarely reported. The present report documents a case of a retroperitoneal lymphoma with double IVC, and discusses its embryological, clinical and radiological significance. In addition, previous cases of a double IVC are reviewed. In the present report, a 52-year-old male patient was hospitalized for a retroperitoneal lymphoma tumor and double IVC. CT urography was performed, whilst a three-dimensional visualization model was also established based on CT data, to reveal a retroperitoneal tumor with double IVC. The present case involved a double IVC with interiliac vein, which was type 2b from the left IVC. The retroperitoneal tumor was identified to be a lymphoma measuring 116x83 mm by percutaneous puncture biopsy. Surgical treatment is generally not recommended for lymphoma. Therefore, this patient was transferred to the Hematology Department for treatment according to the lymphoma management guidelines. The size of the tumor was reduced after chemotherapy during the patient's follow-up. In conclusion, the three-dimensional visualization model can directly and accurately present the anatomical features of the double IVC and its surrounding tissue structure. In addition, variations in the features of IVC can have important clinical significance. It is also important for surgeons, interventional radiologists and clinicians to understand such abnormalities in anatomical features to avoid misdiagnosis and reduce the occurrence of serious intraoperative complications.

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Abbreviations: IVC, inferior vena cava; CT, computed tomography; AA, abdominal aorta; RIVC, right inferior vena cava; 3D, three-dimensional; LIVC, left inferior vena cava; LRV, left renal vein

Key words: double inferior vena cava, anatomical variation, retroperitoneal tumor, three-dimensional reconstruction

Introduction

The inferior vena cava (IVC) is the largest single vein in the human body, and is responsible for collecting venous blood from the abdomen, pelvic organs and lower extremities (1). Because of the complexity of its development during the embryonic stage, IVC can have a variety of anatomical variations in adulthood (2,3). Double IVC is a relatively rare congenital malformation, with a reported incidence of 0.2-3% worldwide (4). Although congenital double IVC is asymptomatic in the majority of cases and it is occasionally found during imaging examination, intraoperative examination or autopsy, this venous malformation may have important implications during surgery and for interventional radiotherapy (1,5-26). Aljabri *et al* (27) reported that fatal and uncontrollable bleeding occurred in 10% of the patients when IVC anomalies were not identified preoperatively. The presence of double IVC can complicate surgery for aortic aneurysm (28). It is important for urologists to evaluate whether the kidney donor has double IVC during the procedure of kidney harvesting, which can prevent vascular injury and complications (29). In addition, patients with double IVC have a high tendency for thromboembolic events (6). The IVC malformations are associated with 5% of deep vein thrombosis cases due to slow blood flow (30). It is crucial to confirm whether double IVC is present for patients requiring the placement of an IVC filter, and venography should be performed to rule out vascular variation for patient with planned IVC filter placement (31). If double IVC is found, the filter should be implanted in both IVCs respectively or a common IVC which is formed by the confluence of the left IVC (LIVC) and right IVC (RIVC), and failure to do so maybe lead to the pulmonary embolism (32). To the best of the authors' knowledge, coexistence of a retroperitoneal tumor with double IVC is rarely reported. In the present report, a case of a retroperitoneal lymphoma with double IVC is documented, following which its embryological, clinical and radiological significance is discussed.

Case report

A 52-year-old male patient of Chinese ethnicity visited the Urology Department of Yichang Central People's Hospital with a retroperitoneal tumor in March 2022, which was discovered unexpectedly during a routine health screen by abdominal ultrasound. The patient was asymptomatic and previously healthy. The patient was assessed using an abdominal computed tomography (CT) for further diagnosis, which revealed a retroperitoneal neoplastic space-occupying lesion measuring 116x83 mm. Furthermore, CT urography was performed using 64-slice spiral CT. The diagnosis was as follows: Right retroperitoneal neoplasm surrounding the right renal artery and vein, abdominal aorta (AA) and RIVC, unclear boundary with the psoas muscle and double IVC (Fig. 1A and C). CT data were collected in the non-enhancement phase, arterial phase and venous phase. The data were then imported into the three-dimensional (3D) reconstruction software (3D visualization image processing software for medical diagnosis, version number: DX3D/V1.4.0. Anhui King Star Digital S&T Co., Ltd.) to establish a 3D visualization model of the abdominal organ tissues and the vascular system to study the overall structure of the double IVC and its relationship with the retroperitoneal tumor. A percutaneous puncture biopsy of the retroperitoneal tumor was then performed. The biopsy tissue of the retroperitoneal tumor was fixed using 10% formalin at 4˚C for 24 h, rinsed with tap water and dehydrated in an ascending series of ethanol followed by xylene. The specimens were then infiltrated and embedded in paraffin, before being affixed to glass slides after sectioning (5 μm). Finally, the specimens were subjected to heating at 37˚C for 12 h, and hematoxylin-eosin staining at 30˚C for 5 min and 3 min respectively. The tumor was then identified to be a lymphoma using light microscopy. The histopathological images showed that fibrous connective tissue was infiltrated by numerous small, round and blue cells (Fig. 2). The patient was transferred to the Hematology Department of Yichang Central People's Hospital in April 2022 for chemotherapy according to lymphoma management guidelines (33). He received 4 cycles of ABVD (doxorubicin, bleomycin, vinblastine, dacarbazine) chemotherapy, following which the size of the tumor was reduced by 3 cm after chemotherapy during the patient's 6 months follow-up.

Abdominal CT showed two rounded structures on both sides of the AA, which were considered to be the bilateral IVC (Fig. 1B). 3D reconstruction showed the bilateral IVC ascending along either side of the AA, where the two vessels had a similar diameter. In front of the left sacroiliac joint, the LIVC appeared to be formed by the confluence of the left internal iliac vein and the external iliac vein. Behind the left common iliac artery, the LIVC rose along the left-side AA to the level of the second lumbar vertebra, ending at the left renal vein (LRV), which crossed anteriorly to the aorta in a normal manner to join the RIVC at an angle of 61˚ (Fig. 3A). The lengths of the LIVC and LRV were 127.8 and 76.7 mm, respectively, whereas the length of the LRV that crosses anteriorly to the aorta was 43.4 mm. The initial caliber of the LRV and left testicular vein was 9.6 and 2.8 mm, respectively, where the blood flows into the LRV (Fig. 3). In front of the right sacroiliac joint, the RIVC was formed by the confluence of the right internal iliac vein and external iliac vein. Behind the right common iliac artery, the RIVC rose along the right side of the AA and joined the LRV to form a common IVC. The right renal vein had a length of 46.9 mm and a caliber of 5.6 mm, which flowed into the common IVC. The length of the RIVC was 142.7 mm, whereas the calibers of the LIVC and RIVC were 14.6 and 14.0 mm, respectively. The caliber of the common IVC was 18.6 mm (Fig. 3A). The retroperitoneal tumor was 116.8x83.9x50.3 mm in size, which tightly enveloped and compressed the RIVC, AA and right renal artery, in addition to pressing the right psoas muscle and bilateral renal veins and the RIVC wrapped (where the tumor was compressing the IVC) was 66.2 mm in length. From the beginning of the common IVC to the level of the RIVC at the right kidney lower pole, no blood supply could be found in the IVC of the wrapped segment. The interiliac vein connecting the LIVC and RIVC had a length of 37.8 mm and a caliber of 6.4 mm. The interiliac vein was located in front of the left sacroiliac joint, crossed posterior to the right common iliac artery, oblique from the lower left to the upper right and joined the RIVC in front of the fifth lumbar vertebra (Fig. 3). According to the classification method of IVC proposed by Chen *et al* (8), the present case involved a double IVC with interiliac vein, which was type 2b from the LIVC.

In the present study, the 3D visualization model was used as a novel diagnostic method. Compared with the 2D CT images, the 3D visualization model provided a stereoscopic and additional detail to the anatomical hierarchy, which was more appropriate compared with that 2D images used in usual clinical practice to meet the patient's clinical requirements. Therefore, these diagnostic procedures were approved by The Ethics Committees of Yichang Central People's Hospital. Written informed consent was obtained from the patient for the participation in the study and publication of this case report.

The literature was then searched. The databases used were https://www.tsgyun.com/official/index.html (wisdom cloud library) and PubMed. The search terms included inferior vena cava, double inferior vena cava, retroperitoneal tumor, anatomical variations, congenital malformation, right inferior vena cava, left inferior vena cava, interiliac vein, three-dimensional visualization model and three-dimensional reconstruction. The inclusion criteria of selecting papers for Table I were double inferior vena cava, baseline characteristics such as sex, age, department in which double inferior vena cava was found, double inferior vena cava diameter, interiliac vein, diagnostic methods, primary symptoms, population and ethnicity and the number of items ≥7 among the 9 items. The exclusion criteria for selecting papers for Table I were minors (<16 years old), article size is less than one page and article is too old (over 30 years).

Discussion

Lucas (34) reported the first case of congenital IVC duplication in 1916. IVC malformations are rare, with the most common type being double IVC, occurring mainly due to abnormalities during embryonic development (25). The venous system originates from three symmetrically paired veins, namely the cardinal veins, umbilical veins and vitelline veins, all of which are formed during weeks 1-4 of embryonic development (26).

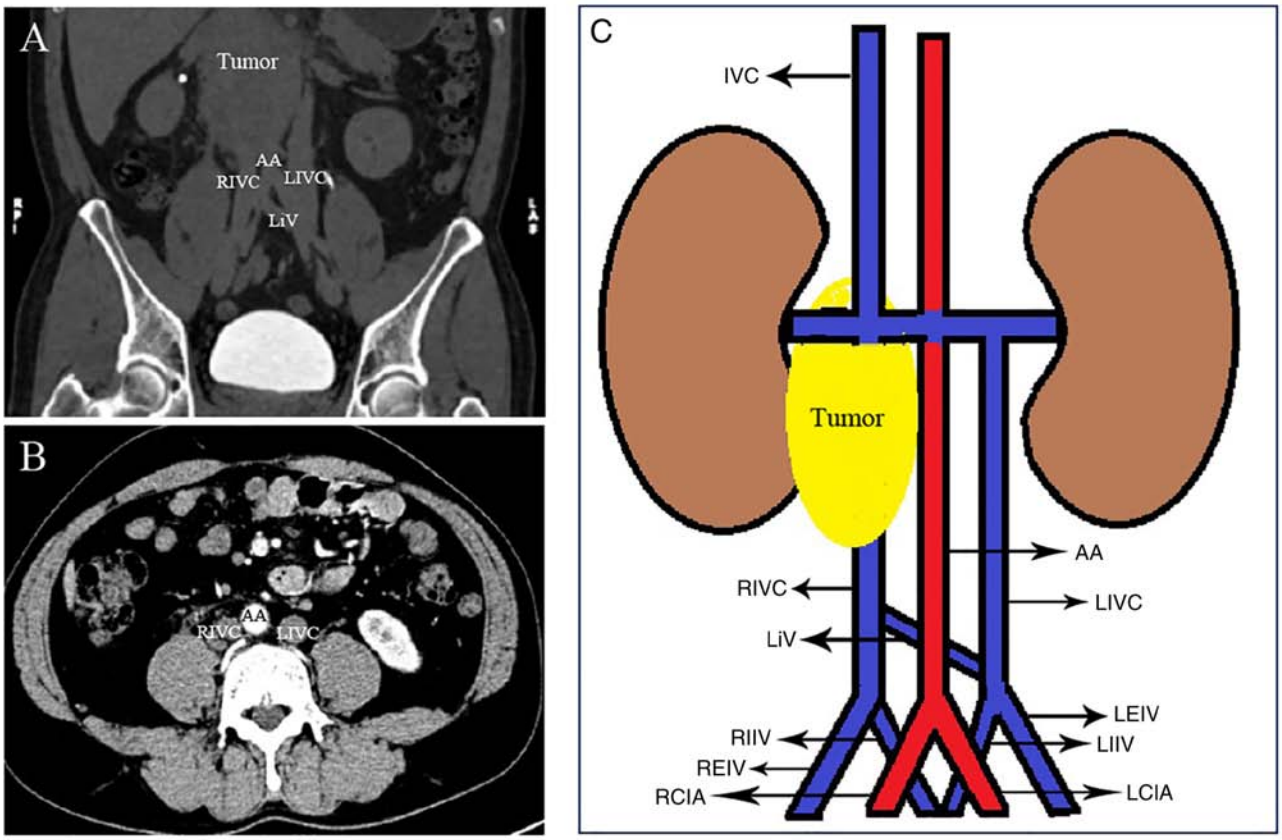

Figure 1. Images of the retroperitoneal lymphoma and double IVC in a 52-year-old male patient. (A) Coronal CT scan showing the double IVC and the retroperitoneal tumor. (B) Axial CT scan showing the double IVC. (C) Schematic showing the double IVC and the retroperitoneal tumor, with the left IVC ending in the left renal vein. AA, abdominal aorta; RCIA, right common iliac artery; LCIA, left common iliac artery; IVC, inferior vena cava; RIVC, right inferior vena cava; REIV, right external iliac vein; RIIV, right internal iliac vein; LIVC, left inferior vena cava; LEIV, left external iliac vein; LIIV, left internal iliac vein; LiV, interiliac vein.

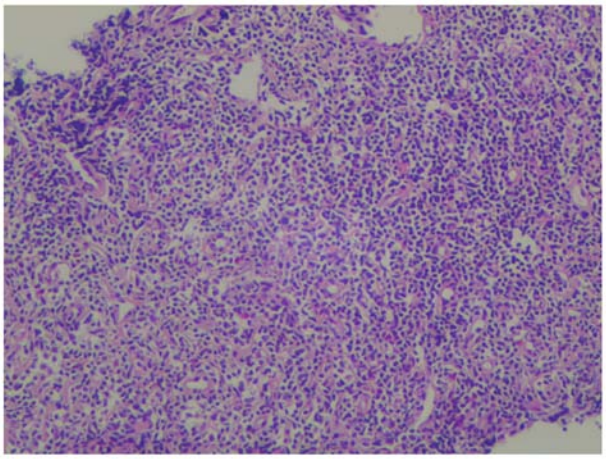

Figure 2. Histopathological image of the retroperitoneal mass. Fibrous connective tissue was infiltrated by numerous small, round and blue cells. H&E. Magnification, x200.

In addition, the IVC develops from four of the following different embryonic sources: The posterior cardinal vein, right subcardinal vein, right supracardinal vein and the right vitelline vein (35). Embryonic development of the IVC is a complex process, involving the formation, anastomosis, regression and replacement of the major embryonic veins (24). The normal IVC consists of the following four parts: Hepatic, renal, suprarenal and infrarenal segments. The hepatic segment was considered to be derived from the vitelline vein (36). By contrast, the suprarenal and infrarenal segments are developed from the right subcardinal vein and right supracardinal vein, respectively (35). The renal segment originates from the right suprasubcardinal anastomosis (36). Left supracardinal vein regression disappears during embryonic development, whereas the right supracardinal vein is retained and develops to form a unilateral right normal infrarenal segment of the IVC (37). Double IVC is caused by the persistence of the bilateral supracardinal veins (37,38).

Our case was a type 2b from the LIVC, and the incidence of type 2b in abnormal IVC has been reported to be 38.5%, where IVC with iliac vein accounts for 67.9% of the total number of cases of IVC abnormalities (8).

The majority of cases of duplicated IVC are asymptomatic (Table I), the diagnosis of which is typically made with CT angiography or MRI. In addition, venography may be used to identify this abnormality. However, there are certain limitations in CT examination. Sousa Gomes *et al* (26) previously reported that a double IVC was found during gynecological surgery, but preoperative CT failed to diagnose this anomaly. In addition, a number of studies have shown that the incidence of double IVC reported based on CT is 0.3-1%, because one of the double IVC may be too narrow, below the scope of detection to be detected with CT (36).

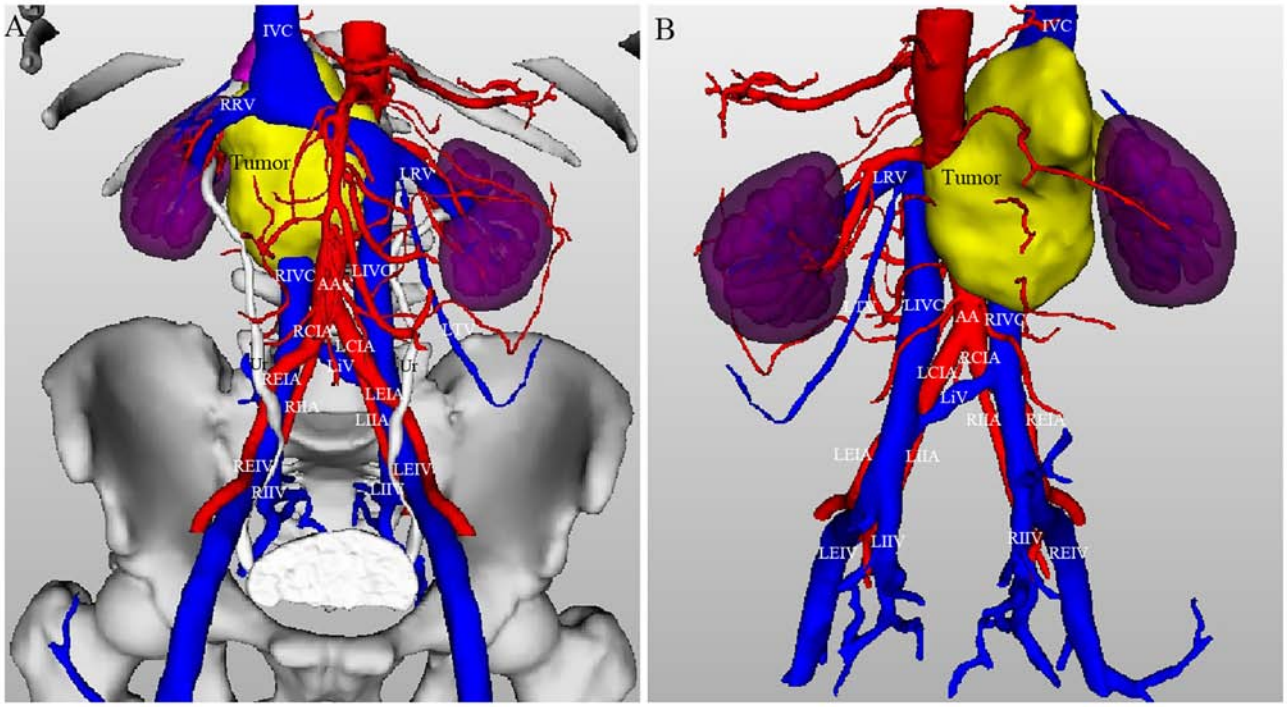

Figure 3. Three-dimensional reconstruction model of the double inferior vena cava and the retroperitoneal tumor based on CT data. (A) Front view. (B) Posterior view. AA, abdominal aorta; RCIA, right common iliac artery; REIA, right external iliac artery; RIIA, right internal iliac artery; LCIA, left common iliac artery; LEIA, left external iliac artery; LIIA, left internal iliac artery; IVC, inferior vena cava; RIVC, right inferior vena cava; REIV, right external iliac vein; RIIV, right internal iliac vein; LIVC, left inferior vena cava; LEIV, left external iliac vein; LIIV, left internal iliac vein; LiV, interiliac vein; RRV, right renal vein; LRV, left renal vein; LTV, left testicular vein; Ur, ureter.

It may sometimes be difficult to use CT to distinguish between venous anomalies and lymphadenopathy, where the two circular structures, one on each side of the aorta, may be misinterpreted as retroperitoneal lymphadenopathy (39). Therefore, in the present report, based on the patient's CT data, a 3D model was reconstructed using 3D visualization software, which enabled multistage fusion visualization of the retroperitoneal tumor, blood vessels and organ tissues. Previous reports on double IVC occasionally use 3D reconstruction images (14,25). The present report utilized a 3D reconstruction model of the retroperitoneal lymphoma with double IVC, which to the best of our knowledge, has not been reported previously. Compared with two-dimensional CT images, the 3D reconstruction model can not only potentially provide an understanding of the anatomical structure of each organ and tissue more intuitively, but can also accurately reflect the positional relationship between the double IVC, AA and tumor by employing freely rotating 3D images.

When a malformation of the IVC is involved in radiology, interventional therapy or surgery, knowledge of double IVC and other vascular variants can be used to minimize the risk of intraoperative bleeding, misdiagnosis or life-threatening complications (9,11,16,22,26). A previous study showed that misdiagnosis of a double IVC caused surgical confusion between the LIVC and the left gonadal vein, resulting in the severing of the LIVC during radical nephroureterectomy (24). Because the left gonadal vein develops from the left subcardinal vein, the LIVC runs along the medial side of the left gonadal vein, which increases the risk of misidentification between the left gonadal vein and LIVC (25). To avoid such misdiagnosis, operators are advised to probe the distal vein during the operation to confirm the type of vein.

In addition, since the interiliac vein crosses anterior to the lumbar and sacral regions, it is of particularly high importance to avoid severe bleeding caused by injury to the interiliac vein during anterior lumbar interbody fusion, anterior sacral and retroperitoneal lymphadenectomy (9,40). Injury to the interiliac vein has been reported to cause serious hemorrhage during gynecological oncology surgery (26). There was an interiliac vein in the present case, where the RIVC was completely blocked by the retroperitoneal lymphoma. However, the RIVC drained into the LIVC through shunting of the interiliac vein and the patient did not develop edema of the right lower limb. Although the patient with lymphoma did not undergo surgery, they were transferred to the Hematology Department for chemotherapy. However, for patients who require surgical treatment, it is important to determine the surgical approach, scope and plans, and to ensure surgical safety according to the 3D visualization technology.

The limitation of the present report is that the patient did not undergo surgical treatment. Therefore, the surgical plan and precautions formulated according to the 3D reconstruction model could not be intuitively verified during surgery.

In conclusion, the present report documented a rare case of a retroperitoneal lymphoma with double IVC. The condition was accidentally discovered by CT. A 3D visualization model was established using 3D reconstruction software based on CT data, which was used to accurately reveal the anatomical details of the double IVC and its surrounding tissue structure. The present case suggested the importance of the recognition

Table I. A review of double inferior vena cava cases reported to 2022.

First author, year	Sex	Age, years	Department	RIVC and LIVC caliber, mm	Interiliac vein	Diagnostic methods	Primary presentation/ Symptoms	Population/ Ethnicity	(Refs.)
Yoshimura *et al*, 2022	Male	85	Department of Anatomy	RIVC, 11; LIVC, 7	Yes	Dissection	NA	Japan/Asian	(5)
Shaheen *et al*, 2022	Male	62	Department of Anatomy	RIVC, 16; LIVC, 16	Yes	Dissection	NA	Saudi Arabia/ Caucasian	(5)
Waśniewska *et al*, 2020	Female	42	Department of Radiology and Diagnostic Imaging	RIVC, 15; LIVC, 13	No	Imaging examination (ultrasonography, CT angiography)	Abdominal pain	Poland/Caucasian	(6)
Klinkhachorn *et al*, 2020	Male	66	Department of Anatomy	RIVC, 16; LIVC, 26	No	Dissection	NA	USA/Caucasian	(7)
Chen *et al*, 2012	Female	84	Department of Anatomy	RIVC, 20; LIVC, 9	Yes	Dissection	NA	Japan/Asian	(8)
Matsuoka *et al*, 2018	Female	53	Gynecologic Surgery	NA	No	Imaging examination (CT)	Advanced ovarian cancer	Japan/Asian	(9)
Matsuoka *et al*, 2018	Female	51	Gynecologic Surgery	NA	Yes	Imaging examination (CT)	Advanced ovarian cancer	Japan/Asian	(9)
Onoda *et al*, 2018	Male	74	Cardiovascular Surgery	NA	No	Imaging examination (CT)	Abdominal aortic aneurysm	Japan/Asian	(10)
Wang *et al*, 2014	Male	32	Vascular Surgery	RIVC, 25; LIVC, 7	No	Imaging examination (venogram)	Pain and swelling of the right lower extremity	China/Asian	(11)
Pilichowska *et al*, 2020	Female	25	Department of Transplantation	NA	No	During the organ procurement procedure	Intracranial hemorrhage	Poland/Caucasian	(12)
Ito and Ikeda, 2018	Female	81	Department of Anatomy	RIVC, 15; LIVC, 10	Yes	Dissection	NA	Japan/Asian	(13)
Coco *et al*, 2016	Female	42	Department of Radiology	NA	No	Imaging examination (CT)	Right abdominal pain	Italy/Caucasian	(14)
Chaijaroonkhanarak *et al*, 2017	Female	45	Department of Anatomy	RIVC, 14; LIVC, 7	Yes	Dissection	NA	Thailand/Asian	(15)
Jiang *et al*, 2011	Male	16	Department of Endocrinology	NA	Yes	Imaging examination (CT, vasography)	Headaches and dizziness	China/Asian	(16)
Nakatani *et al*, 2004	Male	40	Department of Urology	NA	No	Imaging examination (CT, venography)	NA	Japan/Asian	(17)
Nakatani *et al*, 2004	Female	44	Department of Urology	NA	No	Imaging examination (CT, renal angiography)	NA	Japan/Asian	(17)
Kumar *et al*, 2008	Female	62	Department of Urology	NA	No	Imaging examination (CT)	Left flank pain	India/Caucasian	(18)
Yano *et al*, 2000	Male	70	Department of Anatomy	RIVC, 15; LIVC, 13	Yes	Dissection	NA	Japan/Asian	(19)
Yano *et al*, 2000	Male	86	Department of Anatomy	RIVC, 15; LIVC, 10	No	Dissection	NA	Japan/Asian	(19)

Table I. Continued.

First author, year	Sex	Age, years	Department	RIVC and LIVC caliber, mm	Interiliac vein	Diagnostic methods	Primary presentation/ Symptoms	Population/ Ethnicity	(Refs.)
Fronek *et al*, 2006	Female	37	Renal Transplant Unit	NA	No	Imaging examination (angiography, CT)	NA	UK/Caucasian	(20)
Kumar *et al*, 2016	Female	70	Cardiac Center	NA	No	Imaging examination (CT, angiography) examination (CT, angiography)	Dyspnea and chest pain	India/Caucasian	(21)
Furutani *et al*, 2020	Female	66	Gastroenterological Surgery	NA	Yes	Imaging examination (CT, angiography)	Rectal cancer and lung metastasis	Japan/Asian	(22)
Habuchi *et al*, 1993	Male	77	Department of Urology	NA	No	Imaging examination (CT, phlebography)	Asymptomatic gross hematuria	Japan/Asian	(23)
Mao *et al*, 2015	Male	63	Department of Urology	RIVC, NA; LIVC, 13	No	Intraoperative examination	Intermittent gross hematuria	China/Asian	(24)
Yamaguchi *et al*, 2021	Male	60	Department of Urology	NA	No	CT, intraoperative examination	Left scrotal enlargement	Japan/Asian	(25)
Gomes *et al*, 2020	Female	45	Department of Gynecology	NA	Yes	Intraoperative examination	NA	Portugal/Caucasian	(26)

LIVC, left inferior vena cava; NA, not available; RIVC, right inferior vena cava.

of IVC abnormalities. In clinical practice, it is critical to perform preoperative evaluation and preparation for IVC variation, which could be associated with surgery outcomes. This variation in IVC may have important clinical implications. It is of the utmost importance for surgeons, interventional radiologists and clinicians to understand the abnormalities in the anatomical features and to avoid misdiagnosis and reduce the occurrence of severe intraoperative complications.

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Availability of data and materials

All data generated or analyzed during this study are included in this published article.

Authors' contributions

WL and LY made substantial contributions to the design of the study, collected clinical information, and drafted the manuscript, ZD and JX conceived the paper's objective and collected the patient's data. ZL and LZ analyzed the data and performed the literature search. WL and ZD confirm the authenticity of all the raw data. All authors read and approved the final manuscript.

Ethics approval and consent to participate

The present case report was approved by the Ethics Committees of Yichang Central People's Hospital (approval no. 18/16.10.2021). Written informed consent was obtained from the patient.

Patient consent for publication

The patient provided written informed consent for the publication of the information.

Competing interests

The authors declare that they have no competing interests.

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