Case Report

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A deadly trap for para-aortic lymph node dissection in patients with horseshoe kidney as a complication: a case report

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Abstract

Horseshoe kidney is a rare congenital renal dysplasia. It is often associated with various anatomical abnormalities, including renal vessel and ureter variability, which increase unpredictable surgical risks. This current report describes the case of a 42-year-old woman diagnosed as having cervical squamous cell carcinoma complicated by horseshoe kidney. She underwent laparoscopic radical hysterectomy, bilateral oophorectomy and lymph node dissection, including dissection of the pelvic, presacral and para-aortic lymph nodes. The surgery was challenging, but no serious complications occurred. Postoperative multi-slice computed tomography angiography confirmed the anatomical variation of the renal location, ureter and renal vessels. To our knowledge, this is the first reported case of cervical carcinoma complicated with horseshoe kidney.

Keywords

Horseshoe kidney, cervical carcinoma, para-aortic lymph node dissection, inferior vena cava, renal vein, renal artery

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Introduction

Para-aortic lymph node dissection is one of the surgical skills required of gynaecological oncologists. The dissection area ranges from the lower edge of the left renal vein to the midpoint level of the iliac artery region, including lymph nodes and fat tissues

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located by the medial bi-ureter, para-aorta and around the inferior vena cava.[1] The operation is challenging because of the large surgical area and the complex relationship to adjacent organs. The surgical complications include injuries to adjacent organs such as the bowel, ureter, blood vessels, lymphatic vessels and nerves.

Horseshoe kidney (HSK) is a rare congenital renal dysplasia with an incidence of approximately 1 case per 500 individuals.[2] It is often associated with various anatomical abnormalities, including renal vessels and ureter variability, which further increase the unpredictable risks for para-aortic lymph node dissection. Therefore, gynaecologists must be vigilant.

This current report describes a recent case of cervical cancer complicated with HSK. Owing to our limited experience, we underestimated the impact of the anatomical variation in HSK on surgery. The surgery was risky, although serious complications were avoided. According to a literature review undertaken by Shanghai Jiao Tong University, no cases of cervical carcinoma complicated with HSK have been reported to date. This case report aims to remind gynaecological oncologists that a full understanding of the characteristics of HSK, comprehensive preoperative imaging and comprehensive preoperative planning are crucial to avoid unnecessary iatrogenic accidents.

Case report

A 42-year-old woman (gravida 2, para 2) was admitted to Karamay Central Hospital, Karamay, Xinjiang Uyghur Autonomous Region, China in October 2016 complaining of bleeding after intercourse for 1 month. Gynaecological examination revealed a 3-cm cervical cauliflower-like neoplasm with active bleeding. Cervical biopsy showed a squamous cell carcinoma with low differentiation. The preoperative computed tomography (CT) image suggested a space-occupying lesion and HSK (Figure 1). The patient's haemoglobin level was 49 g/l due to sustained vaginal bleeding. Bilateral uterine arterial embolization and blood transfusion were performed. The vaginal bleeding was significantly reduced after the arterial embolization. The patient was diagnosed as having stage IIA1 cervical squamous cell carcinoma. The surgical plan included laparoscopic radical hysterectomy, bilateral oophorectomy, and pelvic, presacral and para-aortic lymph node dissection. This case study was approved the Ethical Review Committee of Karamay Central Hospital. Written informed consent was obtained from the patient.

The renal vein was not detected during the para-aortic lymph node dissection. The abdominal aorta was concomitant with a vascular cord of 0.5 cm in diameter (Figure 2). A large mass with a relatively hard texture and smooth surface was identified across the front of the abdominal aorta and inferior vena cava 6 cm from the common iliac artery bifurcation. At first, the mass was considered to be a metastatic or giant lymph node mass.

To avoid ureter damage, the right ureter was dissociated proximally. The ureter was found to extend into the huge mass. Combined with the preoperative CT scan and intraoperative careful identification, the mass was suspected to be HSK, which was confirmed later intraoperatively by urologists. The junction site of the right ureter and mass was identified to be the deformed renal pelvis (Figure 3).

The unknown vascular cord concomitant with the abdominal aorta possibly originated from the kidney. After presacral lymph node dissection, the vascular cord was found to consist of an artery and a vein. The diameter of the vein was 0.2 cm, which was a variant vein derived from the right kidney and merged into the left

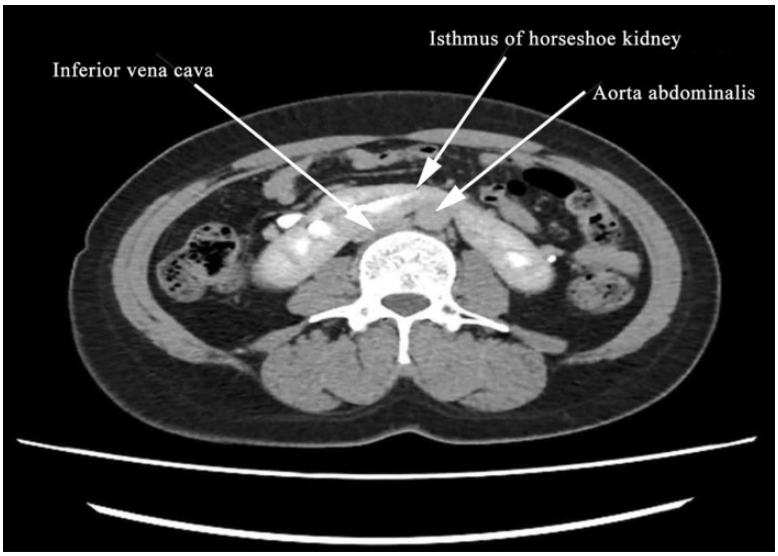

Figure 1. Preoperative computed tomography image of a 42-year-old woman (gravida 2, para 2) who complained of bleeding after intercourse for 1 month. The image was suggestive of a space-occupying lesion and horseshoe kidney.

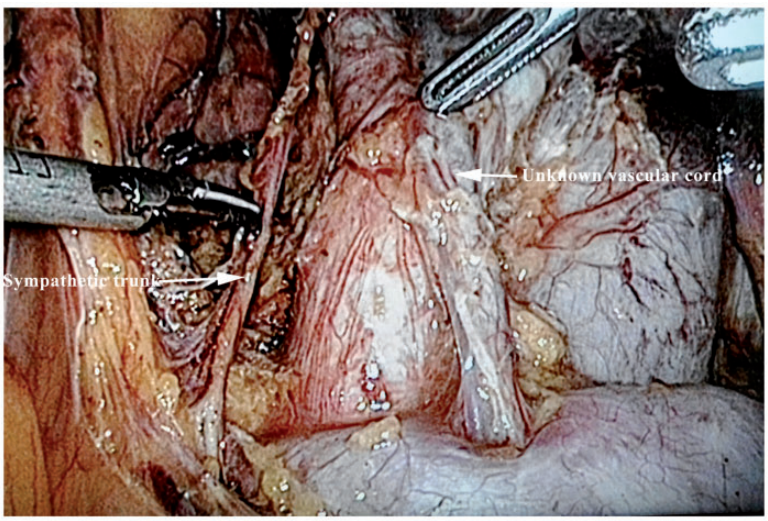

Figure 2. Intraoperative image of a 42-year-old woman who underwent elective surgery for stage IIA1 cervical squamous cell carcinoma showing that the abdominal aorta was concomitant with a vascular cord of 0.5 cm in diameter. The colour version of this figure is available at: http://imr.sagepub.com.

common iliac vein instead of the inferior vena cava. The artery was an aberrant right renal artery derived from the left iliac artery instead of the ipsilateral iliac artery (Figures 4 and 5). The ureter and renal vessels merged into the kidney separately, indicating bilateral renal fusion and external rotation, which was confirmed with

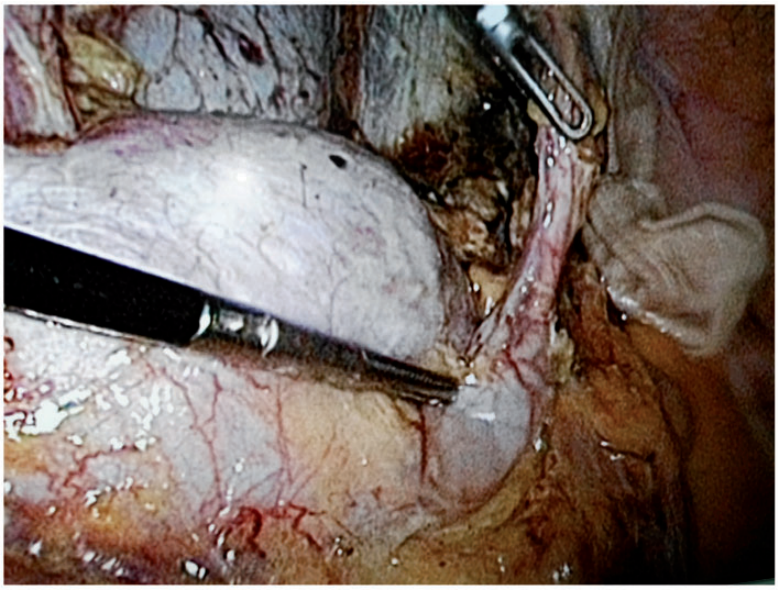

Figure 3. Intraoperative image of a 42-year-old woman who underwent elective surgery for stage IIA1 cervical squamous cell carcinoma. During surgery, a large mass with a relatively hard texture and smooth surface was identified across the front of the abdominal aorta and inferior vena cava. The ureter was found to extend into the huge mass, which was later confirmed to be horseshoe kidney. The junction site of the right ureter and the mass was identified to be the deformed renal pelvis. The colour version of this figure is available at: http://imr.sagepub.com.

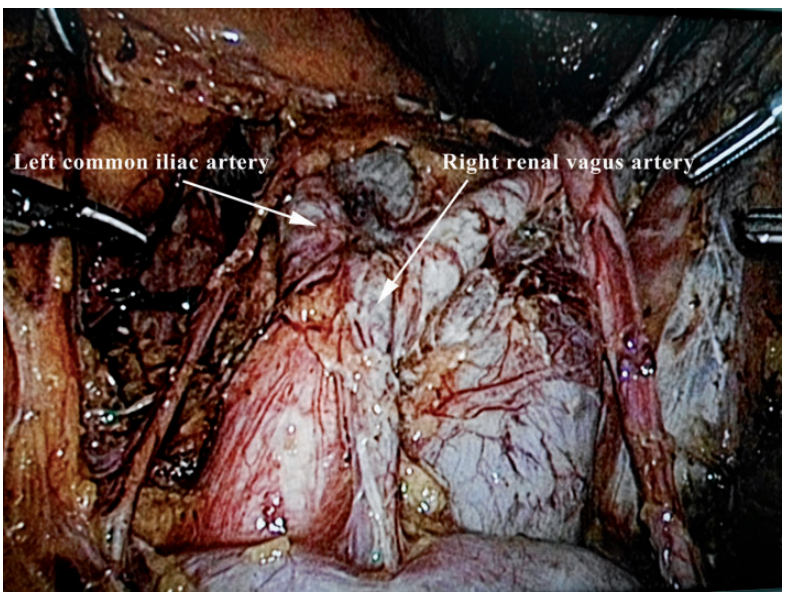

Figure 4. Intraoperative image of a 42-year-old woman who underwent elective surgery for stage IIA1 cervical squamous cell carcinoma showing an aberrant right renal artery derived from the left iliac artery instead of the ipsilateral iliac artery. The colour version of this figure is available at: http://imr.sagepub.com.

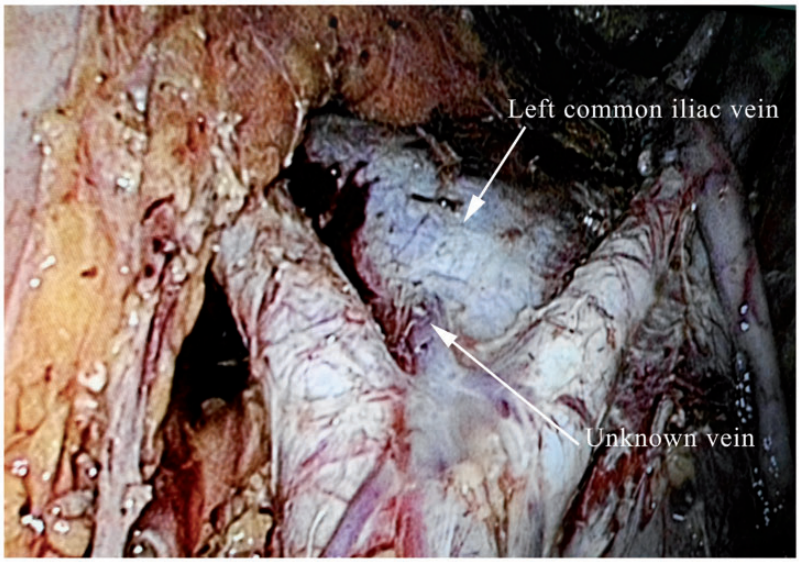

Figure 5. Intraoperative image of a 42-year-old woman who underwent elective surgery for stage IIA1 cervical squamous cell carcinoma showing that an abnormal venous anatomy could merge into the vena cava or internal iliac vein. The colour version of this figure is available at: http://imr.sagepub.com.

postoperative multi-slice computed tomography angiography (MSCTA; Figures 6 and 7).

Pathological examination revealed a highly differentiated cervical squamous cell carcinoma $35 \times 31 \times 25$ mm in size. The cancer had invaded deeply into the muscle layer close to the outer membrane. Local tissues in the cervical inferior lip and vaginal vault were involved. An intravascular cancer embolus was spotted in peri-cancer tissue. The para-aortic, pelvic and presacral lymph modes showed no involvement. The patient was transferred to the radiotherapy and chemotherapy department 2 weeks after the operation. Regular follow-up showed that the patient was generally in good condition with no recurrence or metastasis.

Discussion

In 1895, Emil Ries first established the basic theory of retroperitoneal lymph node dissection and first described the surgical details in 1897.[3] However, the theory remained controversial with regard to the necessity for retroperitoneal lymph node dissection for a long period until wider application of surgical pathological staging of endometrial carcinoma was introduced by the International Federation of Gynaecology and Obstetrics in 1988.[4] At the beginning of the 1950s, Chinese surgeons began to practise radical resection for cervical cancer. Currently, retroperitoneal lymph node excision is an important strategy for cervical and endometrial cancer surgeries. A lymphadenopathy could be suspected when there is CT scan evidence of lymph nodes that are 2 cm in diameter or when there is an intraoperative palpable mass observed during the surgical procedure. Even in this situation, a histological evaluation is necessary because of a high false positive rate of up to 60%.[5] Lymphadenectomy should be performed when a lymphadenopathy is suspected,

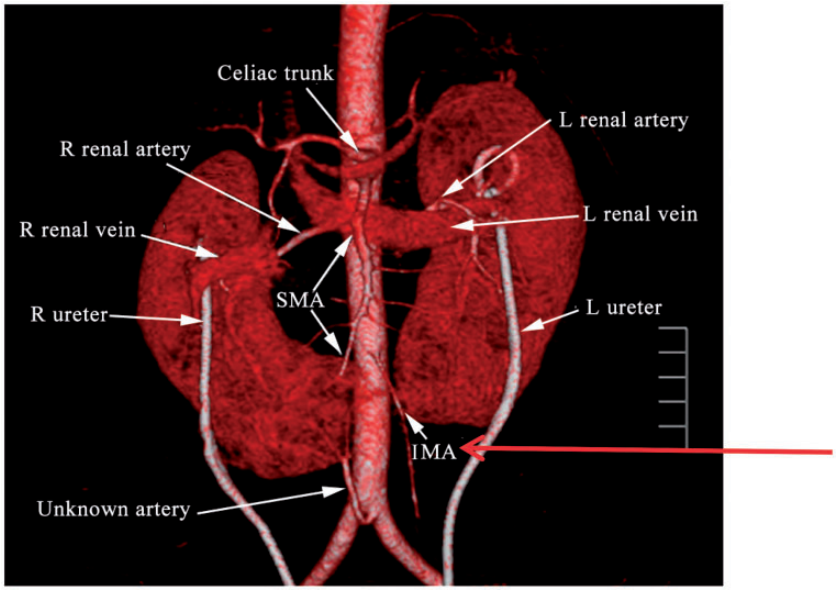

Figure 6. Postoperative multi-slice computed tomography angiography image of a 42-year-old woman who underwent elective surgery for stage IIA1 cervical squamous cell carcinoma confirming that the ureter and renal vessels merged into the kidney separately. The colour version of this figure is available at: http://imr.sagepub.com. R, right; L, left; SMA, superior mesenteric artery; IMA, inferior mesenteric artery.

because it could increase the chances of abrogating the tumour and decrease the risk recurrence.[5] In 1992, the first case of laparoscopic radical resection with hysterectomy, and pelvic and para-aortic lymph node dissection for cervical cancer, was reported.[6]

Owing to the large surgical area and the complex relationship with adjacent organs in para-aortic lymph node dissection, the operation is challenging. If surgeons are not familiar with the anatomical relationship between the adjacent organs in the surgical area, the operation could become dangerous. The incidence of postoperative complications is reported to be 1.0–7.6%.[7,8] HSK often occurs in combination with various anatomical abnormalities involving renal vessels, the ureter, and even the kidney itself, which add unpredictable risks for surgical procedures.[2] Gynaecologists should be vigilant of such cases.

Horseshoe kidney is a congenital kidney fusion deformity.[2] As a rare congenital renal dysplasia, its incidence rate is approximately 1 case per 500 individuals, occurring at any age.[2] The male-to-female ratio is 2:1.[2] Fusion of the kidney bud occurs in the fourth to sixth gestational weeks, which affects renal cephalic migration and the normal rotation process.[2] Patients with HSK are usually asymptomatic if there is no accompanying disease.[9] At least one-third of cases were discovered accidentally.[9] Patients with HSK are more prone to complications of the urinary system compared with healthy controls.[10] The common complications of HSK include renal parenchymal lesions, urinary tract infections, stones and hydronephrosis.[11] The incidence of urinary calculi in HSK patients is as high as 60%, the incidence of obstructive hydrops is 22–40% and the incidence of vesicoureteral reflux is 50%.[10] Abnormal renal fusion

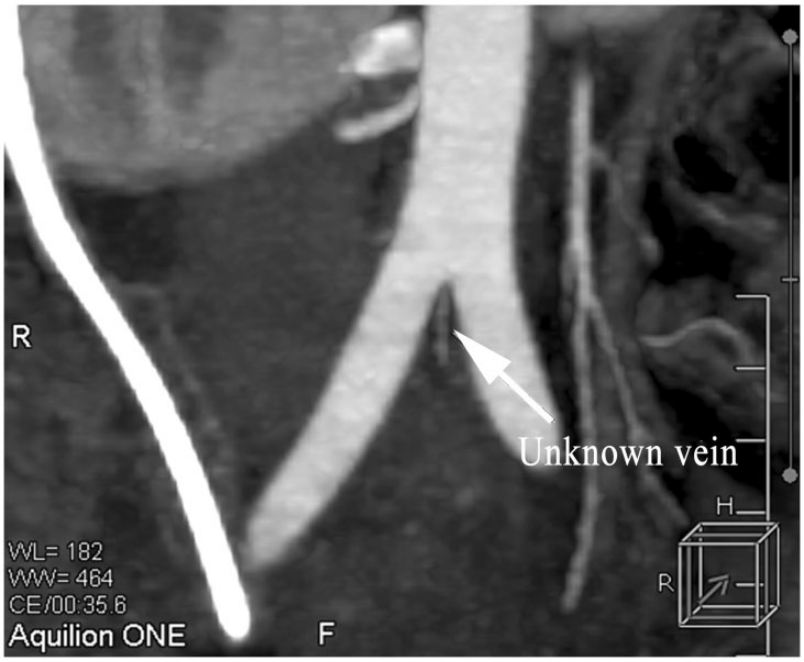

Figure 7. Postoperative multi-slice computed tomography angiography image of a 42-year-old woman who underwent elective surgery for stage IIA1 cervical squamous cell carcinoma confirming that the ureter and renal vessels merged into the kidney separately.

malformation, ureteral distortion and ureteral junction malformation are important factors for complications.[10] The diagnosis of HSK mainly depends on an imaging examination, including ultrasound, intravenous pyelogram (IVP), CT, magnetic resonance imaging (MRI) and other imaging methods.[9] The isthmus connecting the left and right kidneys is the most important direct evidence for the diagnosis of HSK.[12] However, it is easily missed on ultrasound or IVP if the isthmus is fibrous. Therefore, CT and MRI are the recommended methods for diagnosing HSK.[11] CT and MRI can clearly distinguish the renal parenchyma or fibre connection of the isthmus, and the relationship with the surrounding organs.[9] With the development of new imaging technology, multi-slice computed tomography (MSCT) was developed and it can clearly display the abnormal connection of the isthmus, blood supply artery and kidney complications.[13] MSCT can simultaneously display the renal parenchyma, renal collecting system, ureter and bladder. Therefore, MSCT is superior to other diagnostic methods in the classification of abnormal blood supply arteries.[13]

The normal kidneys are located at L1–L2, while HSK is often located below the normal position, at L3–L5, or in the pelvic cavity in 20% of patients, as the process of upward kidney migration during embryonic development is obstructed by the inferior mesenteric artery.[2] The fused part of the HSK is called the isthmus, which is generally located in front of the abdominal aorta and inferior vena cava, but can also rarely be located in the rear of the vessel and even between the blood vessels.[2] In this current case, the

isthmus was located in front of the vessels, at the L3 level (Figure 1). During preoperative planning, the impact of HSK on surgery was underestimated due to limited experience. Therefore, the location of the kidney was ignored. Initially, the mass was considered to be a metastatic or giant lymph node mass rather than an HSK. During surgery, a mass was identified across the front of the abdominal aorta and inferior vena cava at 6 cm from the common iliac artery bifurcation. The first impression of the mass was that of a tumour metastasis. The mass was identified to be in the isthmus until the ureter was dissociated proximally and found to extend into the huge mass. The plan to resect the 'mass' was then abandoned. Our experience in this current case suggests that identification of the kidney location is important for proper surgical planning. Two references for localization should be considered, namely: (i) the inferior edge of the kidney at the lumbar level; and (ii) the relationships of the isthmus location to the abdominal aorta and inferior vena cava.

Horseshoe kidney is often complicated with malrotation, which leads to abnormal ureter location.[2] The ureter in most HSK cases often passes over the isthmus.[2] In some cases, it runs along the anterior surface of the kidney.[2] Cases with the ureter behind the isthmus are rare.[10] In 6% of previously reported cases, duplicate ureters were found at one or two sides of the kidney.[9] These anatomical abnormalities increase the risk of ureteral injury.[9] Preoperative evaluation of ureteral variation, including its number, location and relationship with the isthmus, could help surgeons reduce injury risks. In the present case, the bilateral ureters were located in front of the psoas. Instead of accompanying renal vessels into the renal hilum in the normal anatomy, the bilateral ureter merged into the renal pelvis from the lateral kidney directly, indicating a malrotation of the HSK (Figure 3).

In HSK, a variable vascular supply is a surgical challenge. Usually during renal surgery, a retroperitoneal approach allows early identification of the artery.[14] In this current case, a meticulous study of the imaging data allowed us to salvage the HSK vascular supply. A previous study reported 63% of cases had more than three renal arteries, while single or double renal arteries accounted for 37%.[15] The artery commonly originates from the abdominal aorta, iliac artery and inferior mesenteric artery; but less commonly, it may originate from the internal iliac, external iliac, middle sacral, lumbar and diaphragm arteries.[2,15] HSK can be classified according to six vascular types:[16] type Ia, a renal artery supplies the superior and inferior poles of the bilateral kidneys; type Ib, an artery from the abdominal aorta supplies the bilateral inferior pole of the kidney; type Ic, the bilateral inferior pole of the kidney shares a branch from the abdominal aorta; type Id, multiple renal arteries supply the bilateral kidneys; type Ie, an aortic branch supplies the isthmus; type If, the inferior pole of the kidney is supplied by a branch originating from the mesenteric, common iliac or internal iliac artery. The Graves classification was simplified and no longer describes the supply of contralateral renal arteries (occurring in approximately 25% of cases).[16] A previous study reported that vascular types Ie (28%) and If (24%) were the most common types found in HSK cases.[15] In the present case, the right kidney was supplied by the right vagus artery derived from the left iliac artery, instead of the right iliac artery (Figure 4), which could not be typed using the Graves classification. After peritoneal exposure, a vessel parallel to the abdominal aorta was spotted in front of the aorta. Its source or targeted organs were uncertain; until the confirmation of HSK, it was

considered an aberrant vessel of the kidney. Fortunately, this vessel was not cut off.

Horseshoe kidney is also often accompanied by inferior vena cava abnormalities such as double, left and pre-isthmic inferior vena cava, and the incidence of an anomalous inferior vena cava in patients with HSK is 5.7%.[15] The renal vein usually merges into the inferior vena cava. A previous study reported renal vein variation in 22.9% of HSK patients.[17] An abnormal venous anatomy could merge into the vena cava or internal iliac vein. The current patient's right renal vein was consistent with this variation (Figure 5). Damage to various veins during para-aortic lymph node resection is dangerous in gynaecological cancer surgery. Gynaecologists should be aware of the variability of veins and their branches to avoid accidental intraoperative injury and haemorrhage. Previously, HSK was mainly reported by urologists and seldom reported by gynaecological oncologists. With the increasing frequency of para-aortic lymph node dissection in gynaecological surgeries, gynaecological oncologists should pay attention to organic anatomy variation above the pelvis, such as HSK.

Preoperative imaging is important for understanding the anatomical and vascular variations in patients with HSK. Careful imaging review could help to avoid iatrogenic injuries. Kidney, isthmus, ureter, artery and vein variations should be fully investigated. CT and MRI are of great value for preoperative assessment. CT can accurately display the structure of the HSK and its relationship with the surrounding organs, while enhanced CT scanning can identify the composition of the isthmus. In addition, MSCTA is highly recommended owing to its great advantage in identifying vascular morphology, origin, distribution and relationships between other vascular variations by three-dimensional reconstruction, helping surgeons to fully assess the risks and make appropriate treatment plans.[15] Retrograde pyelography is another choice for visualizing the ureter and kidney with radiography, which might help surgeons to confirm the ureter variability.

In our opinion, the following recommendations should reduce surgical risk during laparoscopic radical hysterectomy, bilateral oophorectomy and lymph node dissection: (i) confirmation of the indication for para-aortic lymph node dissection, performed when indicated; (ii) appropriate preoperative imaging assessments such as CT, enhanced CT, MRI, MSCTA and retrograde pyelography; (iii) appropriate preoperative surgical plan prepared by a multidisciplinary team that includes a urologist and an imaging specialist; (iv) asking for intraoperative assistance when necessary.

Authors' contributions

Qian Zhou drafted the manuscript, reviewed the literature and participated in the operation. Qian Zhou and Xinliang Chen made the histopathological diagnosis. Xinliang Chen performed the operation (surgeon) and revised the manuscript.

Declaration of conflicting interest

The authors declare that there are no conflicts of interest.

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