CASE REPORT

Catheterization in a patient with end-stage renal disease through persistent left superior vena cava: a rare case report and literature review

Huisi He1†, Bingyang Li1†, Yiyi Ma2†, Yuqiang Zhang2, Chaoyang Ye2, Changlin Mei2, Shengqiang Yu2, Bing Dai2* and Yawei Liu2*

Abstract

Background: Persistent left superior vena cava (PLSVC) is a common vena cava malformation, and drains blood into the right atrium via the dilated coronary sinus in most cases. It is usually asymptomatic and detected incidentally during invasive procedures or imaging. Whether the hemodialysis catheters can be placed in PLSVC is still controversial now (Stylianou et al. Hemodial Int 11:42-45, 2007).

Case presentation: Here we report a rare case of catheterization through PLSVC in an end-stage renal disease (ESRD) male patient whose PLSVC connected with pulmonary vein with insufficient blood flow eventually. Among the other 28 cases included in the literature review, 16 cases were non-tunneled catheter and 12 cases were cuffed, tunneled catheter and most of them could provide adequate blood flow.

Conclusion: PLSVC is a rare malformation and mostly asymptotic, we believe that PLSVC drains blood into the right atrium with enough inner diameter and blood flow can serve as an alternative site for conventional dialysis access. However, the feasibility of hemodialysis catheterization through it and measures to avoid serious complications are still needed to be discussed.

Keywords: Persistent left superior vena cava, Hemodialysis catheter, Case report

Background

Persistent left superior vena cava (PLSVC), known as the residual left superior vena cava, is the most common type of vena cava malformations despite its low incidence. In most cases, PLSVC is clinically asymptomatic due to the lack of hemodynamic abnormalities and is almost always found in invasive procedures or imaging.

Reliable and high-quality vascular access which can provide adequate extracorporeal blood flow is a prerequisite for hemodialysis and serves as a crucial factor for prognosis. Non-cuffed and cuffed, tunneled central venous hemodialysis catheter are both preferred choices for end-stage renal disease (ESRD) patients who have an urgent need for hemodialysis, especially when arteriovenous fistula or graft are both unavailable.

The presence of PLSVC brings difficulties and risks for central venous catheterization. Whether the hemodialysis catheters can be placed in PLSVC is controversial until now. Here we report a rare case of hemodialysis catheterization in a patient with ESRD through PLSVC, but it ended with insufficient blood flow compared to the previous case reports.

Case presentations

A 54-year-old hemodialysis patient with a history of multiple central venous catheterizations, arteriovenous fistula, and graft operations was admitted to our unit for the creation of permanent vascular access. After initial screening, an arteriovenous fistula (AVF)/arteriovenous graft (AVG) was deemed not possible due to exhausted...
vasculature of both arms, and a cuffed, tunneled hemodialysis catheter was optioned to be chosen. The right internal jugular vein (IJV) catheterization was attempted under sterile conditions, but the guide-wire could not be advanced more than 10 cm, and the right IJV catheterization was abandoned due to consideration of potential critical stenosis. The left IJV was catheterized with a cuffed, tunneled hemodialysis catheter (14.5F, 36 cm, Palindrome) thereafter without any complication.

Postoperative chest radiograph showed that the catheter was descending straight through the left border of the mediastinum (Fig. 1). Further computed tomography angiography (CTA) of central veins after removal of the hemodialysis catheter, with three-dimensional reconstruction of vessels, revealed the initial segment of the left IJV was stenosed and an abnormal vessel on the left of the aorta drained blood into the left atrium via pulmonary vein. The vascular malformation of PLSVC was confirmed (Fig. 2).

Finally, we replaced a cuffed, tunneled catheter through the right IJV after DSA-guided balloon dilatation of right brachiocephalic venous stenosis. It was removed due to decreasing blood flow and catheter-related bloodstream infection 3 years later. Thereafter, a new cuffed, tunneled catheter was placed in the left IJV which went through right superior vena cava into the right atrium under digital subtraction angiography (DSA) (Fig. 3). Until now, this patient has conducted hemodialysis through the catheter with blood flow around 300 mL/min for 4 years.

**Discussion and conclusion**

PLSVC is the most common kind of congenital malformations in the thoracic vessels. It was first reported by Edwards et al. [1] in 1950 and the latest studies show that the incidence of this deformity is about 0.1–0.5% of the total population, [2] of which about 10% of patients with congenital heart abnormalities [3, 4].

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**Fig. 1** Postoperative chest radiograph showed the location of the cuffed, tunneled hemodialysis catheter and its abnormal path

**Fig. 2** CTA of central vein and three-dimensional reconstruction confirmed PLSVC which connected with pulmonary vein

**Fig. 3** The chest radiograph of the cuffed, tunneled hemodialysis catheter used now.
Human left superior vena cava originates in the third week of the embryonic period, and then the left anterior cardinal vena cava gradually atrophies with embryonic development and finally degenerates into the ligament of Marshall. If the degeneration is not complete, then the remains of a pipeline structure after birth is PLSVC. Some clinicians advocate that it associates with chromosomal aberration, congenital cardiac defect, and extracardiac anomalies might be detected at follow-up [5].

| Types | Characteristics |
|-------|----------------|
| I     | Normal superior vena cava anatomy |
| II    | Only PLSVC exists, without the right superior vena cava |
| IIIa  | PLSVC and the right superior vena cava exist, with left brachiocephalic vein between both sides |
| IIIb  | PLSVC and the right side of the superior vena cava, without left brachiocephalic vein between both sides |

Schummer [6] raised the most recognized classification of the supracardial venous system according to anatomic relationships of superior vena cava and its adjacent (Table 1, Fig. 4). The patient in our case had a type IIIa venous malformation.

Ninety-two percent of PLSVC patients drain blood into the right atrium via the dilated coronary sinus, [7] most of them are asymptomatic and have no hemodynamic abnormalities. In most cases, it's hard to be detected by physical examination and it is always noticed accidentally during imaging or the process of intravascular invasive procedure such as pacemaker implantation, PICC, cardiac electrophysiological examination and central venous hemodialysis catheterization. However, some patients still show abnormal sinus rhythm or bradycardia at the very beginning. In these cases, the patients might undergo pacemaker implantation because of sick sinus syndrome resulting from histological abnormalities caused by an enlarged...
coronary sinus [8, 9]. Another 8% of patients drain blood into left atrium may have obvious clinical cyanosis due to the left to right shunt, and those people always suffer from septal defect, ventricular septal defect or other cardiovascular malformations [10, 11]. This patient’s PLSVC drains blood into the left atrium via pulmonary vein (Type D in Zhu’s classification of PLSVC), but he doesn’t have congenital heart disease and cyanosis which may result from low shunt flow volume (Table 2, Fig. 5) [12].

Can persistent left superior vena cava be used in the hemodialysis catheterization? After a careful literature review, totally 28 cases with hemodialysis catheterization through PLSVC were reported. The details of case reports with hemodialysis catheterization through PLSVC are shown in Table 3. Among them, 16 cases were non-tunneled catheter and 12 cases were cuffed, tunneled catheter. Most of them were type III PLSVC with indwelling catheters in left IJV. The previous history of pacemaker implantation was also notable in the latest case we reported [39]. Among these cases, most operations were completed safely, and hemodialysis catheters met the needs of hemodialysis during the maximum 32-month dwelling time. There was one case reported severe hypotension, bradycardia, and cardiac-respiratory arrest after three times successful hemodialysis. Although the correlation between catheterization and arrhythmia was uncertain, the catheter was removed after the fourth hemodialysis was performed [17]. In another case, rare complication pericardial effusion and bilateral pleural effusions were confirmed by chest computed tomogram since short of breath developed 24 h after catheterization and hemodialysis. This catheter was removed by the cardiothoracic surgeon for safety [28]. In a recently released case, stagnation of blood flow and thrombus formation was found due to a large catheter caliber-to-vein ratio, which resulted in catheter removal after 4 h [38]. Our case is the first hemodialysis patient with PLSVC that drains blood into the left atrium via pulmonary vein, which leads to insufficient blood flow after catheterization. From this rare case and previously reported cases, we raise some concerns about catheterization in PLSVC.

Firstly, the operators should raise awareness of cardiovascular abnormalities during the central venous access.

**Table 2** Zhu’s classification of PLSVC

| Types | Characteristics |
|-------|-----------------|
| A     | PLSVC drains blood to right atrium via coronary sinus |
| B     | PLSVC drains blood to right atrium via coronary sinus with partial right-to-left shunt |
| C     | PLSVC drains blood to left atrium directly with right-to-left shunt |
| D     | PLSVC is directly connected to left pulmonary vein (coronary sinus absent) |

**Fig. 5** Zhu’s classification of PLSVC in dorsal view (SVC: Superior vena cava PLSVC: Persistent left superior vena cava CS: Coronary sinus LA: Left atrium IVC: Inferior vena cava)
| Authors & Year    | Study type | Patient's Gender & Age | The reason of catheterization | The type of SVC | The type of catheter | Catheter Function & Blood Flow (mL/min) | The duration of catheterization | The outcome of catheter or patient | Intraoperative & postoperative complications | Additional anatomical variations |
|------------------|------------|------------------------|-------------------------------|----------------|---------------------|----------------------------------------|---------------------------------|-----------------------------------|--------------------------------------|-----------------------------------------------|
| Kim et al., 1999 [13] | Letter to editor | 28, male | ESRD | Type IIIb | Non-tunneled (left SCV) | Good, 200 mL/min | Unclear but carried out 3 times | Catheter was removed when AVF matured | Not observed | Not observed |
| Pauiter et al., 1999 [14] | Case report | 83, male | ESRD due to DM and HTN | Clear | Non-tunneled (left UV) | Good, Unclear | Unclear | Unclear | Not observed | Not observed |
| Radovic et al., 2002 [15] | Letter to editor | 31, female | ESRD due to | Clear | Non-tunneled (left UV) | Good, Unclear | Unclear | Unclear | Not observed | Not observed |
| Non-tunneled (left UV) | Good, 220 mL/min | 4 weeks | Catheter was removed when AVG was cannulated | Clear | Not observed | Not observed | | | | |
| De la Prada et al., 2002 [16] | Case Report | 45, male | ESRD due to DM | Type III (a or b) | Cuffed, tunneled (right IJV) | Good, 250 mL/min | More than 3 months | Unclear | Not observed | Not observed |
| Diorison et al., 2003 [17] | Case report | 61, female | ESRD due to DM | Type IIIb | Cuffed, tunneled (left UV) | Good, Unclear | Unclear but carried out 4 times | Catheter was removed because of severe arrhythmia | Severe hypotension and bradycardia, cardiac-respiratory arrest | A solitary pelvic kidney |
| Kuppusamy et al., 2004 [18] | Case report | 75, female | AKI due to ischemic tubular necrosis | Type IIIb | Non-tunneled (left UV) | Good, Unclear | Unclear | Unclear | Not observed | Not observed |
| Stylianou et al., 2007 [19] | Case report | 80, female | ESRD due to DM | Type III (a or b) | Non-tunneled (left UV) | Good, Unclear | 1 month | Catheter was removed when AVG was cannulated | Not observed | Anomalous pulmonary vein drainage |
| Orija et al., 2009 [20] | Case report | 72, male | ESRD | Type III (a or b) | Cuffed, tunneled (right IJV) | Good, Unclear | Unclear | Unclear | Not observed | Not observed |
| Parreira et al., 2009 [21] | Case report | 50, unclear | ESRD | Unclear | Cuffed, tunneled (left UV) | Good, Unclear | Unclear | Unclear | Not observed | Not observed |
| Jang et al., 2009 [22] | Case report | 68, male | ESRD | Unclear | Non-tunneled (left UV) | Good, 230 mL/min | Unclear | Unclear | Not observed | Not observed |
| Lim et al., 2010 [23] | Case report | 58, male | ESRD due to DM | Unclear | Cuffed, tunneled (left UV) | Good, Unclear | 5 months | Catheter was removed when AVF matured | Not observed | Aortic coarctation |
| Sriramnaveen et al., 2010 [24] | Letter to editor | 50, male | ESRD due to HTN | Type IIIa | Non-tunneled (left UV) | Good, Unclear | Unclear | Unclear | Not observed | Not observed |
| Authors & Year | Study type | Patient’s Gender & Age | The reason of catheterization | The type of SVC | The type of catheter | Catheter Function & Blood Flow (ml/min) | The duration of catheterization | The outcome of catheter or patient | Intraoperative & postoperative complications | Additional anatomical variations |
|---------------|------------|------------------------|------------------------------|----------------|---------------------|----------------------------------------|-------------------------------|----------------------------------|-------------------------------------|----------------------------------|
| Messina et al., 2011 [25] | Case report | Unclear | ESRD with complete obstruction of central venous vessels | Type III (a or b) | Cuffed, tunneled (left IJV) | Good, Unclear | 15 months | Catheter was replaced with a longer one at 12 months | Not observed | Not observed |
| Kute et al., 2011 [26] | Case report | 45, female | ESRD due to DM and HTN | Type III (a or b) | Cuffed, tunneled (left UV) | Good, 250 mL/min | 2 months | Catheter was removed when AVF matured | Not observed | Not observed |
| Wong et al., 2013 [27] | Case report | Unclear, male | ESRD due to systemic lupus erythematosus | Type IIIa | Non-tunneled (left UV) | Good, Unclear | 3 months | Patient died of pancytopenia and infective endocarditis | Not observed | Not observed |
| Balarubramanian et al., 2014 [28] | Case report | 57, male | AKI | Unclear | Non-tunneled (left UV) | Good, Unclear | 4 h | Catheter was removed by cardiothoracic surgeon | Breathlessness, bilateral pleural effusions, subcutaneous emphysema, pericardial effusion | Not observed |
| Lui et al., 2014 [29] | Case report | 61, male | ESRD due to DM | Unclear | Cuffed, tunneled (left UV) | Good, Unclear | 6 months | Catheter was removed when AVF matured | Not observed | Not observed |
| Kukavica et al., 2014 [30] | Letter to editor | 71, male | ESRD | Unclear | Non-tunneled (left UV) | Good, Unclear | 4 months | Patient died of cerebrovascular stroke, cardio-respiratory insufficiency and cardiac arrest | The failed first two insertions and mild initial resistance during the third insertion | Not observed |
| Dubey et al., 2014 [31] | Letter to editor | 35, male | ESRD (waiting for another renal transplantation) | Type II | Non-tunneled (right UV) | Good, Unclear | Unclear | Unclear | Not observed | Not observed |
| Jaffer et al., 2015 [32] | Case report | 58, female | AKI due to acute tubular necrosis | Type IIIa | Cuffed Tunneled (right UV) | Good, Unclear | Unclear | Unclear | Not observed | Horseshoe kidney |
| Sahutoglu et al., 2016 [33] | Case reports | 80, male | ESRD (acute peritonitis due to peritoneal dialysis) | Type II | Non-tunneled (left UV) | Good, 300–350 mL/min | 3 months | Catheter was removed when AVF matured | Not observed | Not observed |
| | | 35, male | ESRD due to DM and HTN | Type II | Non-tunneled (Right UV) | Good, 300–350 mL/min | 2 months | Catheter was removed when AVF matured | Not observed | Not observed |
| Zhou et al., 2016 [34] | Case report | 63, female | ESRD | Unclear | Cuffed, tunneled (left UV) | Good, Unclear | 9 months | Unclear | Not observed | Not observed |
| Authors & Year            | Study type | Patient’s Gender & Age | The reason of catheterization | The type of SVC | The type of catheter | Catheter Function & Blood Flow (mL/min) | The duration of catheterization | The outcome of catheter or patient | Intraoperative & postoperative complications | Additional anatomical variations |
|--------------------------|------------|------------------------|-------------------------------|-----------------|---------------------|----------------------------------------|----------------------------------|-------------------------------------|-------------------------------------------|-----------------------------------|
| Ricciardi et al., 2017 [35] | Case report | 33, female            | ESRD                           | Unclear         | Cuffed, tunneled (left IJV) | Good, Unclear                          | 32 months                       | Unclear                              | Not observed                             | Cleft lip and palate, uterus bicornis, congenital left hip dislocation and a left inferior vena cava |
| Boodhun et al., 2018 [36] | Case Report | 28, male              | ESRD                           | Type IIIb       | Non-tunneled (left IJV)  | Good, Unclear                          | Unclear                          | Catheter was removed when permanent left femoral catheter was placed | Not observed                             | Not observed                       |
| Anvesh et al., 2018 [37]  | Case Report | 35, male              | ESRD due to HTN                | Type IIIb       | Non-tunneled (left IJV)  | Good, Unclear                          | Unclear                          | Unclear                              | Not observed                             | Not observed                       |
| Kawasaki et al., 2018 [38] | Case report | 66, female            | ESRD due to DM and HTN         | Unclear         | Non-tunneled (left IJV)  | Removed before use                     | 4 h                               | Thrombus formation in the catheter lumen when removed | Not observed                             | Not observed                       |
| He et al., 2018 [39]      | Case report | 88, female            | ESRD due to HTN                | Type II         | Cuffed, tunneled (right IJV) | Good, 220 mL/min                        | 16 months                        | Patient died of gastrointestinal hemorrhage | Not observed                             | Not observed                       |

ESRD: end-stage renal disease, HTN: hypertension, DM: diabetes, AVF: arteriovenous fistula, SCV: subclavian vein, IJV: internal jugular vein, PLSVC: persistent left superior vena cava
For suspected patients with positive symptoms and signs, echocardiography should perform as soon as possible. The direct signs are the existence of the duct-like structure and the blood flow spectrum in the left upper part of the chest, and the indirect sign is the dilated coronary sinus [40]. In addition, unexplained tricuspid atrial systolic murmur and right atrial enlargement should arouse attention. Localized bullae in front of the mediastinum in chest radiography is an important sign of early screening and echocardiography can be the primary screening method. Cardiac catheterization procedure is the gold standard for the diagnosis of PLSVC. However, its invasiveness, radioactivity prohibits clinical use. Thoracic enhanced CTA might serve as an alternative.

Secondly, left IJV is a preferred cannulation site for hemodialysis catheterization through PLSVC, especially for those patients with absent right superior vena cava. Traditionally, right JIV cannulation is generally preferred in hemodialysis patients due to its straight path directly into the superior vena cava and fewer complications compared with other positions. Nevertheless, in these PLSVC without right superior vena cava cases, since the right IJV and subclavian vein drains blood into PLSVC via the right brachiocephalic vein, traditional right JIV cannulation may encounter difficulties and acute complications normally met in left IJV cannulation. Central vein perforation, pneumothorax, and arterial puncture all have been reported in previous cases, which mostly caused by force during the operation without the sense of cardiovascular malformations. So, whenever any resistance is met with forwarding the guidewire or the peel-away sheath, do not push by force, what you need is to pull it out and reassess vascular condition (especially for PLSVC with absent right superior vena cava). Detailed history survey, preoperative imaging screening, intraoperative fluoroscopic guidance, and postoperative chest radiograph assessment for suspected patients are priority points to avoid serious complications.

Thirdly, whether a hemodialysis catheter can be placed in PLSVC is still controversial until now [19]. Our case proved that the PLSVC which rarely drains blood into the left atrium via pulmonary vein or left-to-right shunt cannot be used to conduct hemodialysis because of obvious hemodynamic abnormalities and insufficient blood flow. In most cases, PLSVC flowed back into the right atrium through the coronary venous sinus. Although few complications were reported in the placement of a non-tunneled hemodialysis catheter through PLSVC (Table 3), hemodynamic changes after indwelling catheters in those patients potentially may lead to angina pectoris, arrhythmia, stroke, cardiac arrest due to coronary sinus irritation. In severe cases, it may threaten the patients’ life [19, 22, 41]. Some nephrologists believe that PLSVC is relatively thin and the blood flow is not enough to maintain long-term hemodialysis, and the locally generated turbulence may increase the probability of thrombosis and arrhythmia. However, if the diameter of PLSVC and blood flow were sufficient, with stably flowed back through the coronary venous sinus into the right atrium, it was feasible to dwell a hemodialysis catheter in PLSVC for long-term hemodialysis. We believe that after an accurate assessment of intrathoracic vessels including the inner diameter of PLSVC via preoperative imaging, a PLSVC can serve as an alternative site for conventional dialysis access.

However, the location of the catheter tip remains to be elucidated. The tip of the cuffed, tunneled hemodialysis catheter is normally positioned within the right atrium or at the junction of superior vena cava and right atrium. For PLSVC patients, the right atrium is inaccessible and the placement of catheter tip in the left superior vena cava that is close to the coronary sinus might cause arrhythmia, so we think that the lower left superior vena cava with adequate blood flow and negative cardiac effect might be an optimal choice.

PLSVC is a rare and asymptotic malformation, so the early detection and diagnosis before hemodialysis catheterization are quite difficult. Detailed history survey, echocardiography and preoperative imaging screening are the priority points to identify suspect patients. Rarely, the PLSVC which drains blood into the left atrium via pulmonary vein or left-to-right shunt should be excluded. During the surgery, intraoperative ultrasound and fluoroscopic guidance are strongly recommended if available. Performing catheterization carefully, position it properly and do not push it by force may help to avoid serious complications. We believe that PLSVC drains blood into the right atrium with enough inner diameter and blood flow can serve as an alternative site for conventional dialysis access. Besides, the preferable location of the catheter’s tip with minor hemodynamic effect remained to be determined.

Abbreviations
AVG, arteriovenous graft; CTA, computed tomography angiography; DSA, digital subtracted angiography; ESRD, end-stage renal disease; IJV, internal jugular vein; PLSVC, persistent left superior vena cava

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Availability of data and materials
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Authors’ contributions
HSH, BYL, YYM, YWL and BD were all directly involved in the care of the patient. HSH and BYL acquired the data necessary for analysis and wrote the initial draft of the paper. YYM, YQZ, CYJ, CLM and SQY contributed in data analysis and interpretation. BD and YWL were responsible for revising the manuscript. All authors approved the final version of the manuscript prior to submission. All authors agreed to be accountable for all aspects of the final manuscript.

Ethics approval and consent to participate
Not applicable.

Consent for publication
The patient received all information regarding this case report. Written informed consent for publication in BMC Nephrology was obtained from the patient.

Competing interests
The authors declare that they have no competing interests.

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Author details
1Naval Clinical Medicine Grade 2014, Basic Medical College, Naval Medical University, Shanghai 200433, People’s Republic of China, 2Department of Nephrology, Changzheng Hospital, Naval Medical University, Shanghai 200433, People’s Republic of China.

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