A New Technique: Laparoscopic Resection of Fibrous Ring and Placing Extravascular Stent in Patients With Nutcracker Syndrome: A Report of 5 Cases

Fang-Min Chen, Xu-Long Chen, Xi-Nan Jiang, Shuai Tang, Jia-Qi Shi, Deng-Bao Li, Si-Wen Zhong, and Yu Chen

OBJECTIVE To evaluate the feasibility and safety of a new laparoscopic technique for resection of a fibrous ring and extravascular stent implantation in patients with nutcracker syndrome (NCS).

MATERIAL AND METHODS We retrospectively reviewed data of 5 patients diagnosed with NCS between March 2010 and February 2016. The mean age of the patients (4 male and 1 female) was 34 years (range, 28-40 years). All 5 patients underwent laparoscopic resection of the narrow fibrous ring around the left renal venous (LRV) and for extravascular stent implantation in the LRV for management of NCS.

RESULTS The average operating time was104 minutes and the average blood loss during surgery was 59 mL. The average length of the postoperative hospital stay was 6 days (range, 4-8 days). In all 5 patients, the symptoms of macroscopic hematuria started decreasing gradually and resolved after surgery. Postoperative computed tomography showed that the blood outflow from the LRV was smooth. The ratio of the dilated segment's inner diameter to the diameter of the strictured segment decreased from 3.4 to 9.5, preoperatively to 1.1-2.0, postoperatively. The mean follow-up period was 17.6 months (range, 8-24 months).One patient's varicocele was cured and symptoms in all 5 patients resolved after surgery. None of the patients showed symptom recurrence.

CONCLUSION Laparoscopic surgery, for the placement of an extravascular stent and resection of the fibrous ring around the end of the LRV outflow to the inferior vena cava appears feasible and safe and offers an alternative minimally invasive for the management of NCS. UROLOGY 126: 110–115, 2019. © 2019 Elsevier Inc.

NCS, also known as the left renal vein (LRV) compression syndrome or nutcracker phenomenon, refers to compression of the LRV between the aorta abdominalis and the superior mesenteric artery (SMA).[1] It can cause symptoms of gross or microscopic hematuria, orthostatic proteinuria, intermittent left flank pain, left varicocele (male), pelvic congestion syndrome (female), and so on.[2,3] The diagnostic procedure of NCS is often difficult and is commonly delayed. The surgical approaches should be considered when symptoms are severe or failure to respond to conservative treatment after 1 year, and current surgical treatment approaches include LRV transposition with patch venoplasty, transposition of the LRV distally to the inferior vena cava (IVC), renal autotransplantation, endovascular stenting, etc.[4] Here we report 5 cases procedure about laparoscopic management of NS with resection of fibrous ring and placing extravascular stent from March 2010 to February 2016.

CASE PRESENTATION

Five cases (4 male, 1 female) were studied. The mean age was 34 years (range, 28-40 years). The average duration of

All authors agree to recommend Fang-min Chen as the joint first author. All authors read and approved the final manuscript.

This work was supported by Science and Technology Fund Project of Health Development Planning Commission of Guizhou Province of China (no. gzwjkj 2014-2-119). The authors declare no competing financial interests. The author Xu-Long Chen contributed to the study equal to first author and he was recommend to be the co-first author of this paper.

Conflict of interests: *The authors declare that they have no competing interests.*

Ethical standard: *This study was approved by the ethics committee of the affiliated hospital of Guizhou Medical University. All patients were informed and consented prior to their data collected in the study. This article contained no personal information of any patients enrolled.*

From the Department of Urology, Tianjin Third Central Hospital, Tianjin, People's Republic of China; the Department of Urology, Affiliated Hospital of Guizhou Medical University, Guiyang, People's Republic of China; and the Department of Medical Record, Tianjin Third Central Hospital, Tianjin, People's Republic of China

Address correspondence to: *Dr. Fang-Min Chen, Department of Urology, Tianjin Third Central Hospital, No.83 Jintang Rd., Hedong District, Tianjin 300171, People's Republic of China. E-mail: cfm_cn@aliyun.com*

Submitted: July 2, 2018, accepted (with revisions): December 31, 2018

https://doi.org/10.1016/j.urology.2018.12.039
0090-4295

clinical symptoms was 35 months (range, 6-72 months). All cases were characterized by recurrent gross hematuria, intermittent left kidney pain without hypertension, oliguria, frequent micturition, dysuria, and fever. On physical examination, the average body mass index was 18.36 kg/m^2, left flank abdominal pain was noted in 4 cases, and left varicocele combined with NCS was noted in 1 male patient with no decrease in the degree of varicosity in the prostrate position. None of the studied cases had urinary tract stones, a previous history of deep vein thrombosis, or any other kidney disease. Urinalysis showed routine red blood cells (3+ to 4+); the red blood cells were of nonglomerular origin, except for 3 cases visible lean body mass proteinuria. Other laboratory results were normal. A regular cystoscopy showed blood oozing from the left ureteric orifice in 3 cases. Preoperative duplex ultrasound scans in all 5 cases revealed a compressed LRV, between the aorta and the SMA, with an average peak velocity of 121.4 cm/s (range, 110-132 cm/s). Computed tomography (CT) showed that the average stricture segment diameter of the LRV was 1.8 mm (range, 1.2-2.5 mm). The average maximum proximal dilatation diameter of the LRV in the renal hilum was 10.5 mm (range, 8.3-15.2 mm). The average ratio of the dilated segment's inner diameter to the strictured segment's inner diameter was 6.1 (range, 3.4-9.5; Table. 1). Preoperative CT examinations for vascular showed a compression of the LRV between the aorta and the SMA in the 5 cases (Fig. 1A). The preoperative diagnosis was LRV compression syndrome. All patient information was reviewed by the hospital ethics committee, and all patients signed the informed consent form.

The patients were placed in the lateral decubitus position under general anesthesia. A standard 4-port transperitoneal technique was used, and the umbilical incision was the first point for pneumoperitoneum access (the CO_2 pressure was maintained at a level about 15 mm Hg). The retroperitoneum in the avascular area of the mesenterium was incised along the inner margin of the descending colon to reach the retroperitoneal cavity. The LRV, IVC and the aorta were exposed successively. Careful dissection was performed to prevent injury to the spleen and venous collaterals. The fibrous bundles between the aorta and SMA were separated to expose the LRV (Fig. 2B). Intraoperatively, the LRV became flat when the SMA was elevated, while the proximal LRV became engorged when the artery was put down. This laparoscopic observation further confirmed the diagnosis of NCS. The decision to preserve or transect the left gonadal vein, adrenal central vein, and lumbar veins was made on a case-by-case basis. A matter of concern was the discovery of a narrow fibrous ring around the LRV outflow end to IVC in all 5 cases. The fibrous ring was resected (Fig. 2C), after which the dilated proximal LRV showed immediate shrinkage (Fig. 2D). An externally reinforced, expanded polytetrafluoro-ethylene graft (Bard Peripheral Vascular, Inc.), 12 mm in diameter, was introduced and bound to the surface of the LRV along the long axis to prevent blood vessel compression. The graft was finally fixed to the surface

Table 1. Treatment outcomes in 5 patients with nutcracker syndrome

Patient	Sex	Age	Symptoms/ Signs	Aortomesenteric Angle	LRV Anteroposterior Diameter (mm), Hilum/Aorto-mesenteric Portion (Supine)	LRV Peak Velocity (cm/s), Aortomesenteric/Hilum Portion (Supine)	Follow-Up (Months)	Outcomes
1	Male	31	Gross hematuria; varicocele	28	15.2/2.5	115	16	Relief
2	Female	28	Gross hematuria; anemia	31	10.8/1.8	110	24	Relief
3	Male	34	Gross hematuria; flank pain	18	9.7/1.6	126	24	Relief
4	Male	40	Gross hematuria; flank pain	22	8.8/1.2	124	8	Relief
5	Male	38	Gross hematuria;	26	8.3/2.0	132	24	Relief

LRV, left renal vein.

Figure 1. Preoperative and Postoperative CT views of NCS. (**A**) Preoperative a compression of the LRV between the aorta and the SMA. (**B**) Postoperative placement of the ES. (**C**) Postoperative 3-dimensional vascular remodeling about relieving of the LRV and the ES. (Color version available online.)

of the aorta to avoid shifting (Fig. 2E). The postoperative Doppler ultrasonography and computed tomography showed that the average blood outflow peak velocity from the LRV decreased to 34 cm/s (range, 25-45 cm/s), whereas the average preoperative peak velocity was 121.4 cm/s (range, 110-132 cm/s).

All patients met the criteria for a definitive diagnosis of NCS. Diagnostic imaging techniques such as ultrasonography and CT revealed the entrapment of the LRV between the SMA and the aorta. Intraoperatively, the entrapment of the LRV between the aorta and the SMA with proximal dilation was clearly visible. No complications occurred during the laparoscopic procedure. The average operation time was 104 minutes and the average blood loss during surgery was 59 mL. The average length of postoperative hospital stay was 6 days. Postoperative Doppler ultrasonography and computed tomography measurements showed that the blood outflow of the LRV was smooth (Fig. 1B), and that the inner diameter had decreased in size (Table 2).The varicocele was cured in 1 case. The average follow-up period was 17.6 months (range, 8-24 months). One male patient continued to have recurrent, but tolerable, flank pain but the cause of this pain was not clear.

DISCUSSION

The anatomical abnormality resulting in compression of the LRV between the abdominal aorta and the SMA was first described as the NCS in 1950.[5] The common clinical symptoms associated with this syndrome include hematuria, flank pain, proteinuria, pelvic congestion in female patients, and left-sided varicocele in male patients.[6] The proteinuria observed in this syndrome can easily be misdiagnosed as an urinary tract infection, nephrolithiasis, papillary necrosis, glomerulo-nephritis, or even retroperitoneal cancer and frequently leads to the misdiagnosis of NCS.[7,8] Recent studies have shown that Schonlein-Henoch purpura IgA nephropathy, membranous nephropathy and hypercalcemia can also occur concurrently with

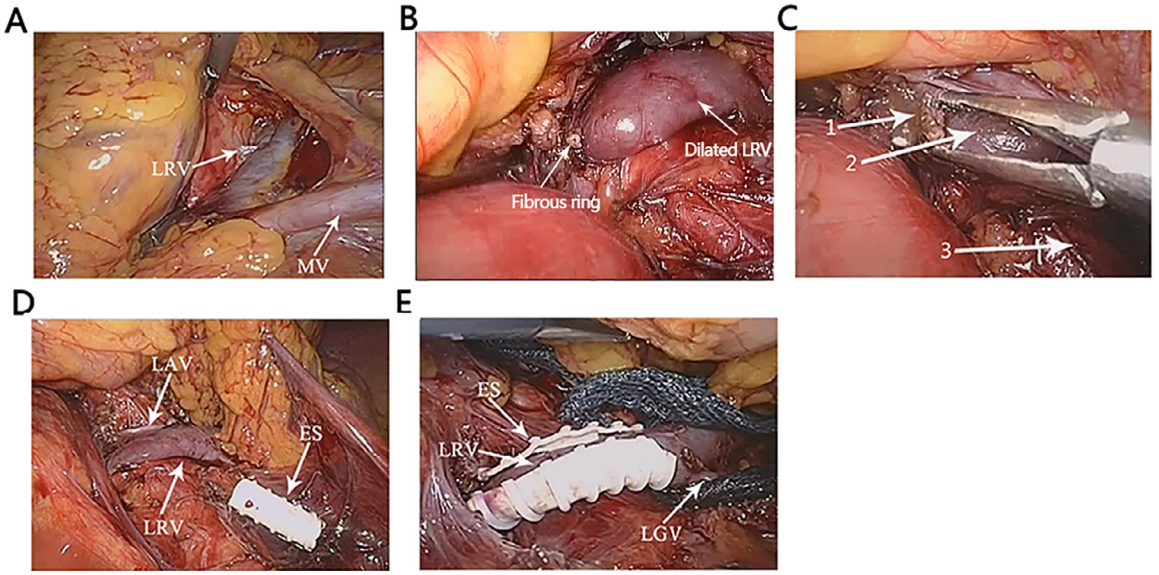

Figure 2. Intraoperative imagings. (**A**) Exposure of the dilated LRV. (**B**) Exposure of the dilated LRV. (**C**) Separation and resection of the fibrous ring around the LRV outflow of the IVC. Pointing to separation of the fibrous ring around the LRV outflow of the IVC, dilated LRV, and aorta abdominalis with the numbers 1, 2, and 3, respectively. (**D**) The shriveled LRV after resection of the fibrous ring around the LRV. (**E**) Placement of the ES. (Color version available online.)

Table 2. Patient's characteristics in 5 cases

Variable	Patient 1	Patient 2	Patient 3	Patient 4	Patient 5
Sex	Male	Female	Male	Male	Male
Age, y	31	28	40	38	34
Symptoms/Sign	Gross hematuria; varicocele	Gross hematuria; anemia; flank pain	Gross hematuria; flank pain	Gross hematuria; flank pain	Gross hematuria; flank pain
Urine cytology	proteinuria	Hematuria	Negative	Hematuria	Hematuria
CT	Entrapment of the LRV	Entrapment of the LRV	Entrapment of the LRV	Entrapment of the LRV	Entrapment of the LRV
Aortomesenteric angle	28	31	18	22	26
Urethrocystoscopy	Negative	Hematuria	Negative	Hematuria	Hematuria
LRV anteroposterior diameter (mm)/aortomesenteric portion (supine)					
Before operation	15.2/2.5	10.8/1.8	9.7/1.6	8.8/1.2	8.3/2.0
Post operation	5/3.9	4.7/3.7	5.9/3.5	5.1/3.2	4.7/3.5
LRV peak velocity(cm/s), portion (supine)					
Before operation	115	110	126	124	132
Post operation	45	36	34	25	30
Left renal arteriogram	Abnormal dilation of the LRV	Abnormal dilation of the LRV	Abnormal dilation of the LRV	Abnormal dilation of the LRV	Abnormal dilation of the LRV
Treatment	LERVS	LERVS	LERVS	LERVS	LERVS
Operative duration, min	75	97	117	81	150
Blood loss, mL	50	70	60	50	65
Surgical drain, d	1	1	1	2	1
Complications	None	None	None	None	None
Discharge, d	5	4	5	6	4
Follow-up	16	24	16	8	24
Outcome	Relief	Relief	Relief	Relief	Relief

CT, computed tomography angiography; LERVS, laparoscopic extravascular renal vein stent placement; LRV, left renal vein.

this condition, and have been implicated in the pathogenesis of NCS.[9] NCS associated symptoms lead to misdiagnoses such as urinary tract infections, kidney stones, and glomerulo-nephritis.[10]

These factors make the diagnosis of NCS challenging. The diagnosis relies on clinical manifestations, duplex ultrasonography, and CT venography. All these techniques are noninvasive and should be widely used. The normal length of the LRV is about 6-10 cm, the diameter is about 4-5 mm, and the normal pressure gradient between the LRV and IVC is less than 1 mm Hg. If the pressure exceeds 2 mm Hg, NCS should be highly suspected.[1,9,11] A pressure gradient >3 mm Hg can be used as the basis for the diagnosis of NCS. CT angiography and magnetic resonance angiography can provide a clear picture of the anatomy and can help estimate the diameter of the blood vessel.[12] The accuracy of CT angiography is 91.7%, with a specificity of 88.9%.[13] Magnetic resonance angiography has a similar diagnostic value. An invasive test such as cystoscopy may be helpful to identify hematuria with a left ureteral origin.[14]

Five patients in this group complained of long-term gross hematuria and underwent color Doppler ultrasound examination. The ratio of the uncompressed LRV section to the narrow strictured LRV section was between 3.4 and 9.5. The CT Image Reconstruction Check showed obvious LRV compression (Fig. 1 A). Endoscopy data in 3 cases showed hematuria with a left ureteral origin. Diagnosis was confirmed based on the previously mentioned criteria. The management of NCS depends upon the clinical presentation and severity of the LRV hypertension. Different management options have been proposed for NCS, including follow-up, conservative treatment, and surgical therapy. These patients, who were less than 18 years of age, with gross hematuria (with no anemia) and thin physique, could be followed up. Some experts believe that the follow-up period should not be less than 2 years for patients younger than 18. During the follow-up period, 75% of patients with hematuria experienced a complete relief from symptoms.

Surgical treatment includes intra- or extravascular stenting and open surgery.[6,15] Open surgery options include LRV transposition, gonadocaval venous bypass, renal autotransplantation, and nephropexy.[16] Minimally invasive management techni ques include both endovascular stent placement and laparoscopic surgery and include various methods such as laparoscopic splenorenal venous bypass, laparoscopic LRV-IVC transposition for NCS, and laparoscopic renal autotransplantation. Although endovascular stent placement is a simple and attractive choice, it may result in fibromuscular hyperplasia and stent embolization or migration, potentially leading to left nephrectomy.[17] However, severe flank pain, renal functional impairment, and inefficacy of or aggravation after conservative treatment for more than 2 years might require surgical treatment.

In this group of 5 cases with severe recurrent gross hematuria, 1 case had severe varicocele and all 5 had experienced a failure of the conservative treatment protocol for more than 2 years. In addition, this group of patients was lean, including 3 patients with a body mass index less than 18.5 kg/m^2. A significant improvement in symptoms was not observed while using a conservative treatment (through measures such as better nutrition) for weight gain. We recommend surgical treatment for underweight NCS patients, if their response to conservative treatment approaches is poor. Traditional surgical methods such as SMA transposition and SMA excision surgery may cause intestinal ischemia, necrosis, anastomotic bleeding, stenosis, and other complications. Renal vein transposition surgery requires kidney isolated in case of a renal artery block, resulting in a greater impact on renal function.[2,6,17] Renal trauma and vascular anastomotic complications are potential complications of autologous transplantation which limit the usefulness of this procedure.

In all the patients in this group, we found a narrow fibrous ring around the end of the left renal venous outflow to the inferior vena cava (Fig. 2B and C). After resection, an extravascular stent was placed around the LRV for the treatment of NCS. We did not dissociate or block the left renal artery around the LRV or SMA at any point during the surgical procedure, having advantages of less trauma, fewer complications, faster recovery, and relieving the LRV coarctation fast. Careful selection of patients for the treatment of NCS with laparoscopic techniques can effectively reduce the risk of vein bypass reconstruction.

We would like to emphasize some key points based on our experience. First, the SMA, the renal vein, and the aorta should be dissected and dissociated carefully and safely. Second, the surgical procedure involves the resection of a narrow fibrous ring from the outflow end of the proximal LRV and IVC junction (Fig. 2B and C). Therefore, protecting the adrenal vein and reproductive vein, while fully exposing the LRV, in preparation for the surgical placement of stent is needed. Third, attention must be paid to the placement of stent without renal artery ligation, to avoid renal warm ischemia injury and vascular injury. Last, the graft was wrapped around the LRV, fixed, and sutured to the myolemma of the erector spinae and adventitia of the abdominal aorta to prevent translocation (Fig. 1B and C).

It is worth mentioning that the technique allowed us to note the existence of a clear fibrous ring around the LRV outflow to the IVC. The fibrous ring could have been an additional cause for compression in addition to the anatomic extrinsic compression on the LRV. Therefore, the laparoscopic procedure should not only be used for treatment of the disease, but also serve to confirm the diagnosis. After the procedure, the dilated region of the LRV showed immediate shrinkage (Fig. 2D). Thus, we postulate that the compression of the LRV in NCS may be related more to the existence of the fibrous ring on the LRV, rather than due to the extrinsic compression of the LRV as it crosses between the SMA and the aorta.

CONCLUSION

Treatment of patients with NCS by laparoscopic resection of a fibrous ring and placement of an extravascular stent around the compressed LRV is quick and safe. The magnification effect in laparoscopy offers excellent visibility, and results in substantially lower blood loss than with open surgery. Moreover, surgical procedures can be completed without blocking the renal artery, thereby avoiding renal warm ischemic injury. Our study shows that this technique is an effective method for the treatment of NCS.

References

1. Hilgard P, Oberholzer K, Meyer zum Buschenfelde KH, et al. The "nutcracker syndrome" of the renal vein (superior mesenteric artery syndrome) as the cause of gastrointestinal complaints. *Dtsch Med Wochenschr*. 1998;123:936–940. https://doi.org/10.1055/s-2007-1024101. (1946).
2. Hartung O, Grisoli D, Boufi M, et al. Endovascular stenting in the treatment of pelvic vein congestion caused by nutcracker syndrome: lessons learned from the first five cases. *J Vasc Surg*. 2005;42:275–280. https://doi.org/10.1016/j.jvs.2005.03.052.
3. Chung BI, Gill IS. Laparoscopic splenorenal venous bypass for nutcracker syndrome. *J Vasc Surg*. 2009;49:1319–1323. https://doi.org/10.1016/j.jvs.2008.11.062.
4. Rogers A, Beech A, Braithwaite B. Transperitoneal laparoscopic left gonadal vein ligation can be the right treatment option for pelvic congestion symptoms secondary to nutcracker syndrome. *Vascular*. 2007;15:238–240.
5. Hartung O, Barthelemy P, Berdah SV, et al. Laparoscopy-assisted left ovarian vein transposition to treat one case of posterior nutcracker syndrome. *Ann Vasc Surg*. 2009;23:413. https://doi.org/10.1016/j.avsg.2008.08.026. e413-416.
6. Hartung O, Azghari A, Barthelemy P, et al. Laparoscopic transposition of the left renal vein into the inferior vena cava for nutcracker syndrome. *J Vasc Surg*. 2010;52:738–741. https://doi.org/10.1016/j.jvs.2010.04.018.
7. Zhang Q, Zhang Y, Lou S, et al. Laparoscopic extravascular renal vein stent placement for nutcracker syndrome. *J Endourol/Endourol Soc*. 2010;24:1631–1635. https://doi.org/10.1089/end.2010.0001.
8. Chen S, Zhang H, Tian L, et al. Endovascular management of nutcracker syndrome after migration of a laparoscopically placed extravascular stent. *Am J Kidney Dis*. 2012;60:322–326. https://doi.org/10.1053/j.ajkd.2012.04.022.
9. Estraviz B, Arevalo JA, Eizaguirre E, et al. Laparoscopic approach for the multiple abdominal vascular entrapment syndrome. *Hepatogastroenterology*. 2012;59:175–177. https://doi.org/10.5754/hge10452.
10. Viriyaroj V, Akranurakkul P, Muyphuag B, et al. Laparoscopic transperitoneal gonadal vein ligation for treatment of pelvic congestion secondary to Nutcracker syndrome: a case report. *J Med Assoc Thai*. 2012;95(Suppl 12):S142–S145.
11. Xu D, Gao Y, Chen J, et al. Laparoscopic inferior mesenteric-gonadal vein bypass for the treatment of nutcracker syndrome. *J Vasc Surg*. 2013;57:1429–1431. https://doi.org/10.1016/j.jvs.2012.10.092.
12. Bisoi AK, Sahu MK, Chander CN, et al. Nutcracker syndrome in a young female with solitary functional left ovary: a surgical challenge. *Ann Vasc Surg*. 2014;28:1938. https://doi.org/10.1016/j.avsg.2014.07.019. e1931-1933.
13. Li P, Shao P, Qin C, et al. Retroperitoneal laparoscopic extravascular stent placement for renal nutcracker syndrome: initial experience. *Urol Int*. 2014;92:396–399. https://doi.org/10.1159/000353349.
14. Chen M, Liu N, Chen S, et al. Laparoscopic inferior mesenteric to gonadal vein end-to-side bypass: a new attempt for nutcracker syndrome treatment. *Ann Vasc Surg*. 2015;29. 1321 e1329-1321 e1311. https://doi.org/10.1016/j.avsg.2015.03.049.
15. Shao P, Li P, Ju X, et al. Retroperitoneal laparoscopic reimplantation of the left renal vein for nutcracker syndrome. *Urol Int*. 2015;94:74–78. https://doi.org/10.1159/000362423.
16. Syed F, Lam Q, Maharjan N, et al. Diagnosis and successful surgical management of posterior nutcracker syndrome in a patient with loin pain hematuria. *J Ark Med Soc*. 2015;111:254–256.
17. Wang SZ, Zhang WX, Meng QJ, et al. Laparoscopic extravascular stent placement for nutcracker syndrome: a report of 13 cases. *J Endourol/Endourol Soc*. 2015;29:1025–1029. https://doi.org/10.1089/end.2014.0411.