## A novel hybrid left renal vein transposition and endovascular stenting technique for the treatment of posterior nutcracker syndrome

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#### ABSTRACT

Posterior nutcracker syndrome occurs when a retroaortic left renal vein becomes compressed between the abdominal aorta and the lumbar spine. Although open surgical approaches remain the treatment of choice, endovascular stenting has been used successfully. We describe a case of a 28-year-old man who presented with microscopic hematuria, left-sided flank pain, and testicular swelling. Computed tomography findings were consistent with posterior nutcracker syndrome. He underwent a novel hybrid operation that included left renal vein transposition followed by endovascular stenting. Repeated imaging at 3 and 12 months revealed a patent stent with complete resolution of symptoms. (J Vasc Surg Cases and Innovative Techniques 2017;3:142-5.)

The left renal vein (LRV) takes a retroaortic course in 0.5% to 3% of the population.<sup>1</sup> Posterior nutcracker syndrome (PNCS) occurs when the patient presents with symptoms related to the compression of this retroaortic LRV (between the aorta and the lumbar spine).<sup>2</sup> This abdominal venous entrapment can lead to venous hypertension, manifested as flank pain, hematuria, varicocele, and ureteropelvic junction obstruction.<sup>2</sup> We describe a case of PNCS treated with a novel hybrid operation that included LRV transposition followed by endovascular stenting. The patient gave consent for the publication of this report.

#### **CASE REPORT**

We describe a 28-year-old man with past medical history significant for cerebral palsy and cerebrovascular accident at birth, now with residual left-sided weakness and scoliosis, who presented with left-sided flank pain and hematuria. The pain had persisted for several weeks with radiation to the left groin and notable testicular swelling. He experienced no dysuria, nocturia, stranguria, or frequency changes. He reported being sexually active with multiple partners and quitting smoking a year before after a 2.5 pack-year smoking history. Urinalysis at the time of

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initial presentation with flank pain was positive for microscopic hematuria. The results of recent tests for human immunodeficiency virus infection, gonorrhea, and chlamydia were negative. Laboratory workup was unremarkable, with a white blood cell count of 8.51  $\times$  1000  $\mu/L$ , hemoglobin concentration of 15.6 g/dL, hematocrit level of 46.5%, blood urea nitrogen concentration of 11 mg/dL, and creatinine concentration of 0.87 mg/dL. Computed tomography venography of the abdomen and pelvis demonstrated a retroaortic LRV (Fig 1), which was also confirmed on vascular ultrasound. Scrotal ultrasound examination demonstrated a left-sided varicocele with vein thrombosis and no right-sided abnormalities. Initially, the patient was observed without any intervention. Hematuria remained stable with no drop in red blood cell counts. However, his flank pain did not improve during the next 8 months. Therefore, operative intervention was offered to the patient. The patient underwent a novel hybrid operation that included open LRV transposition with venoplasty followed by endovascular stenting.

### **OPERATIVE DESCRIPTION**

Initially, inferior venacavography and left renal venography were performed through the right common femoral vein. This demonstrated flattening of the LRV (Fig 2) and reflux of contrast material into the left gonadal vein. A pressure gradient of 1 mm Hg between the LRV and the inferior vena cava (IVC) was measured. Then, through a midline laparotomy, exposure of the aorta, IVC, and retroaortic LRV was obtained. Under systemic heparinization (with an activated clotting time of >225 seconds), multiple tributaries of the LRV were ligated and divided. The LRV was detached from the IVC, and this was closed. The LRV was then transposed anterior to the aorta and reattached at a more distal portion of the IVC. The LRV was sewn in an end-to-side fashion by a venoplasty using bovine pericardium to avoid tension in the anastomosis and to augment the short retroaortic LRV. Once this portion of the operation was completed, we proceeded endovascularly with left

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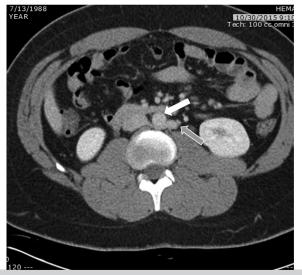
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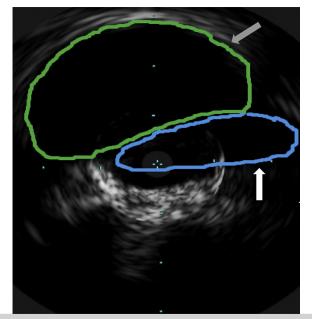
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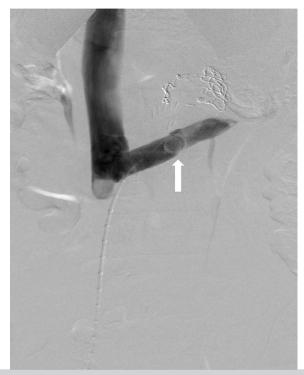


**Fig 1.** Axial view of computed tomography venography demonstrating a retroaortic (*white arrow*) left renal vein (LRV; *gray arrow*).



**Fig 2.** Intravascular ultrasound demonstrating flattened left renal vein (LRV; *white arrow*) lying posterior to the abdominal aorta (*gray arrow*).

renal venography, which allowed sizing of the newly transposed LRV. An 18  $\times$  40-mm self-expanding Wallstent (Boston Scientific, Marlborough, Mass) was placed into the LRV (Fig 3). Subsequently, to prevent migration and embolization of the Wallstent, this stent was sewn into place using interrupted 5-0 Prolene sutures before closure of the abdomen. The postoperative course was uneventful. The patient was started on dual antiplatelet therapy with aspirin 81 mg and clopidogrel 75 mg daily for the first 3 months, which was then switched to aspirin



**Fig 3.** Venogram after placement of self-expanding stent (*arrow*) into the left renal vein (LRV).

325 mg. Repeated computed tomography venography at 3 and 12 months after the operation demonstrated a patent stent. Furthermore, the patient reported resolution of hematuria, flank pain, and testicular edema.

#### DISCUSSION

The treatment of nutcracker syndrome aims to reduce LRV hypertension and subsequent pelvic congestion. This can be achieved through open surgical or endovascular approaches. However, because of the rarity of this anomaly and the lack of long-term follow-up of patients in the endovascular arm, the open surgical option has remained the "gold standard" of therapy. Of the PNCS cases reported since 2013, 5 of 10 were corrected surgically, whereas only 2 were corrected endovascularly (Table).

The open surgical treatment includes LRV transposition with or without venoplasty. Other alternative surgical options, including renocaval bypass, renal autotransplantation, and nephrectomy, have been reported.<sup>3</sup> Complications of the open approach are similar to those of other intra-abdominal procedures, including paralytic ileus, retroperitoneal hematoma or hemorrhage, and persistent pelvic pain. Some reports have shown that patients with lower pressure gradients may experience less impressive results as these gradients may persist after surgery.<sup>4</sup> Hematuria was found to decrease shortly after surgery, recurring only intermittently or with additional trauma.<sup>4</sup>

Table. Reported	cases of posterior	nutcracker syndrome	(PNCS)

Author	Year	No. of patients	Presentation	Treatment
Ozkam et al	2016	1, M, 16 years old	Flank pain	Conservative
Deser et al	2016	1, F, 36 years old	Flank pain, hypertension	Open LRV transposition with polytetrafluoroethylene graft placement
Koh et al	2015	1, M, 24 years old	Hematuria	Conservative
Zhang et al	2015	1, M, 58 years old	Flank pain, hematuria	Open prosthetic left renocaval bypass
Syed et al	2015	1, F, 22 years old	Flank pain, hematuria	Left laparoscopic nephrectomy with autotransplantation
Quinones-Baldrich et al	2015	1, M, 5 years old	Hematuria	Open LRV transposition and LRV aneurysm resection
Chen et al	2014	1, F, 8 years old	Macroscopic hematuria	Open LRV transposition
Allam et al	2014	1, M, 44 years old	Flank pain, hematuria	Endovascular express balloon-expandable 10 $\times$ 37-mm stent
Granata et al	2014	1, M, 22 years old	Microscopic hematuria	Endovascular self-expandable 14 × 40-mm stent
Shah et al	2013	1, F, 46 years old	Hematuria	Conservative
LRV, Left renal vein.				

The use of endovascular techniques in PNCS has largely been extrapolated from their use in the anterior nutcracker syndrome. Quevedo et al<sup>5</sup> reported >150 cases of successful endovascular stenting of anterior nutcracker syndrome with resolution of symptoms. Variable stents from 10 to 20 mm in diameter and 40 to 60 mm in length have been used. Size selection for the stent is highly reliant on precise LRV measurements by intravascular ultrasound before placement, and it is usually oversized up to 20% to prevent migration.<sup>6</sup> The ideal stent should have high radial strength with little shrinkage in length to allow accurate placement.<sup>6</sup> To date, endovascular intervention has been described in only two cases of PNCS that used a 10  $\times$  37-mm balloon-expandable stent and a 14  $\times$  40-mm selfexpandable stent, respectively (Table).<sup>7,8</sup> Both cases were successful at eliminating hematuria and flank pain. However, complications, such as stent thrombosis and erosion, are feared.<sup>6</sup> Although endovascular stenting is a less invasive option, further follow-up specific to the area of PNCS is warranted before it can be deemed a first-line treatment of PNCS.

It has been reported that after undergoing LRV transposition, patients will still have a 20% rate of reintervention in an endovascular fashion because of LRV stenosis or thrombosis.<sup>9</sup> Therefore, we propose this novel approach of immediate stenting at the time of LRV transposition not only to increase the radial strength of the vessel, preventing collapse or thrombosis, but also to avoid technically challenging reintervention at a later date. Furthermore, we think that posterior stent placement without LRV transposition unnecessarily predisposes young patients to the development of an aorto-LRV fistula or potential stent erosion into the spine, and long-term benefit of stenting is lacking. The timing of intervention also remains controversial. It has been proposed that patients younger than 18 years can undergo a 2-year period of conservative therapy, given a high likelihood of spontaneous remission in up to 75% of patients.<sup>2</sup> No such specification is made for symptomatic adult patients. Our patient received surgery at the 8-month mark because of persistent flank pain not amenable to analgesics. Similarly, there are no standard guidelines for antiplatelet or anticoagulation therapy. Most cases report the use of one of these agents for at least 2 to 3 months.<sup>10</sup> Further observation will be required to define ideal surgical vs endovascular candidates.

#### CONCLUSIONS

PNCS remains a rare anomaly that can be effectively relieved when it is symptomatic by a novel combination of surgical transposition and endovascular techniques. This case highlights the importance of recognizing the potential vascular origin of flank pain. A longer followup time and further experience are required to fully evaluate the efficacy and safety of this novel hybrid technique.

#### REFERENCES

- 1. Ayaz S, Ayaz UY. Detection of retroaortic left renal vein and circumaortic left renal vein by PET/CT images to avoid misdiagnosis and support possible surgical procedures. Hell J Nucl Med 2016;19:135-9.
- Skeik N, Gloviczki P, Macedo TA. Posterior nutcracker syndrome. Vasc Endovascular Surg 2011;45:749-55.

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- 3. Zhang X, Wang S, Wei J. Prosthetic left renocaval bypass for posterior nutcracker syndrome. Indian J Surg 2015;77(Suppl 1):103-5.
- 4. Reed NR, Kalra M, Bower TC, Vrtiska TJ, Ricotta JJ 2nd, Gloviczki P. Left renal vein transposition for nutcracker syndrome. J Vasc Surg 2009;49:386-93; discussion: 393-4.
- 5. Quevedo HC, Arain SA, Abi Rafeh N. Systematic review of endovascular therapy for nutcracker syndrome and case presentation. Cardiovasc Revasc Med 2014;15:305-7.
- 6. Policha A, Lamparello P, Sadek M, Berland T, Maldonado T. Endovascular treatment of nutcracker syndrome. Ann Vasc Surg 2016;36:295.e1-7.
- 7. Allam SR, Livingston TS, Kalaria V, Diaz-Calderon W, Pumphrey JA, Patel L, et al. Posterior nutcracker

syndrome: an infrequent cause of hematuria. Kidney Int 2014;85:985-6.

- 8. Granata A, Clementi A, Floccari F, Di Lullo L, Basile A. An unusual case of posterior nutcracker syndrome. Clin Exp Nephrol 2014;18:670-1.
- 9. Erben Y, Gloviczki P, Kalra M, Bjarnason H, Reed NR, Duncan AA, et al. Treatment of nutcracker syndrome with open and endovascular interventions. J Vasc Surg Venous Lymphat Disord 2015;3:389-96.
- 10. Kurklinsky AK, Rooke TW. Nutcracker phenomenon and nutcracker syndrome. Mayo Clin Proc 2010;85:552-9.

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