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Surgical Outcomes of Nutcracker Syndrome: A Single Center Experience

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Abstract

Background: Nutcracker syndrome (NCS) is a rare vascular compression disorder characterized by left renal vein (LRV) entrapment, leading to various clinical manifestations.

Aim of Study: The current study aimed to evaluate the surgical outcomes of patients with NCS.

Material and Methods: A retrospective analysis was conducted on 10 patients with NCS who underwent surgical intervention at Tanta University Hospitals, Egypt between January 2021 and February 2024. Intraoperative techniques employed varied based on individual findings, including LRV transposition, patch venoplasty, and renocaval bypass grafting.

Results: The mean age of the patients was 20.3±3.09 years, with a predominance of males (80%). The mean body mass index was 20.5±2.96kg/m^2, and the mean aortomesenteric angle was 24.3±4.14°. Postoperatively, a significant reduction in pain (p<0.001) and hematuria (p=0.010) was observed compared to preoperative levels. Varicocele showed an insignificant difference (60% vs 20%, p=0.169). Improvement was noted in 80% of patients, while 20% experienced postoperative infections.

Conclusions: Surgical intervention for NCS demonstrated promising outcomes, with significant alleviation of pain and hematuria in the studied cohort. While varicocele improvement was statistically insignificant, the majority of patients experienced overall symptomatic relief.

Key Words: Nutcracker syndrome – Surgical outcomes – Transposition – Venoplasty – Renocaval bypass.

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Introduction

THE nutcracker syndrome (NCS) is an uncommon vascular compression disorder characterized by signs or symptomatology arising from left renal vein (LRV) compression within the aortomesenteric space resulting in proximal dilation of the LRV **[1]**.

The NCS has two subtypes: Anterior nutcracker syndrome (ANCS) and posterior nutcracker syndrome (PNCS). ANCS causes LRV compression due to congenital or acquired narrowing of the aortomesenteric space, often co-occurring with Wilkie's syndrome **[2]**. Left-sided varicocele is linked to 9% of patients with retroaortic left renal vein (RLRV) and nutcracker syndrome, with renal collar or circum-aortic venous ring causing anomalies **[3]**.

In the "renal collar" condition, the presence of two left renal veins (LRVs) – one coursing anterior and the other posterior to the abdominal aorta – predisposes individuals to "combined nutcracker syndrome." This occurs due to the potential for concomitant compression of both the anterior LRV (situated between the aorta and superior mesenteric artery) and the posterior LRV (located between the aorta and spine) **[4]**.

In addition to LRV entrapment can be caused by extrinsic compression, intrinsic valvular incompetence, and elevated venous pressure. Posterior NCS involves compressed LRV between aorta and vertebral column **[5]**. Compression of the renal venous system leads to elevated venous pressure, causing varicosities, haematuria, orthostatic proteinuria, and pain symptoms like left flank pain, abdominal pain, pelvic pain, dyspareunia, dysmenorrhea, and fatigue **[6]**.

The NCS presents complex clinical presentations, lack of specific diagnostic criteria, and heterogeneity in patient symptoms, making definitive diagnosis challenging. Females may develop pelvic congestion syndrome, while males may experience left varicocele formation **[7].**

The NCS diagnosis is primarily done through imaging studies, with Doppler ultrasonography being the most common. Treatment strategies vary based on patient severity, with conservative management for younger patients and surgical interventions for severe cases **[5].**

Treatment approach may be conservative in the presence of tolerable symptoms, and surgical or hybrid and stenting procedures in the order of priority in the presence of intolerable symptoms **[8].**

The NCS described as an "uncommon vascular compression disorder," implying that it is a relatively rare condition. Conducting studies on rare conditions is essential to expand the knowledge base and improve patient management. Therefore, the current study aimed to evaluate the surgical outcomes of patients with NCS treated at Tanta University Hospitals.

Patients and Methods

This retrospective study involved 10 patients with NCS aged ≥18 years of either sex with NCS characterized by symptoms of macroscopic and microscopic hematuria, proteinuria, and flank pain who failed to improve on conservative treatment admitted to Tanta University Hospitals between January 2021 and February 2024. Approval obtained from the Institutional Ethical Committee at Faculty of Medicine, Tanta University, Federal Wide Assurance with Approval Code: 36264PR407/11/23.

All patients provided written informed consent.

Exclusion Criteria were patients with significant missing data.

Outcome measures:

- Pain is assessed by comparing the visual analogue scale preoperatively and postoperatively.
- Hematuria is assessed by laboratory examination of RBCs in urine.
- Varicocele is assessed clinically and by duplex ultrasound

Preoperative assessment:

All patients underwent a preoperative assessment including detailed history (age, sex, body mass index), complete physical examination, and review of routine laboratory investigations (complete blood count, coagulation profile, renal function tests, liver function tests, electrolytes). Vital signs (blood pressure, temperature, heart rate, respiratory rate) were also recorded. Computed tomography (CT), cross-sectional imaging (magnetic resonance venography or CT venography) (Fig. 1), and duplex ultrasound were performed to evaluate the anatomy and hemodynamics related to NCS. Venography with LRV catheterization assessed patency and pressure gradients.

Intraoperative techniques:

Following exploratory laparotomy, through midline incision, the specific surgical approach depended on intraoperative findings. Potential techniques include: 1- Removal of fibro-lymphatic bands compressing the renal vein to ensure sufficient aorto-mesenteric angle 2- Transposition of LRV in front of SMA (Fig. 2) 3- External synthetic graft cuff shield around the left renal vein using ringed PTFE double 8mm synthetic graft (Fig. 3), 4- LRV transposition to the inferior vena cava (Fig. 4) 5- Left gonadal vein bypass to left Common iliac vein (Fig. 5). The surgical steps involved mobilization of the LRV and dissection of the aorto-mesenteric window with removal of all adhesions. The choice of the operative technique depends upon the anatomy at the compressed part. If there is no segment of LRV to the right side of the SMA towards the IVC, the decision was resection of the LRV from the IVC with closure of the venotomy of IVC by lateral venorrhaphy then re-implantation of the LRV to the IVC by tension-free end-to-side anastomosis to the lateral IVC few centimeters caudal to the original location. If there's a segment of the LRV available segment of LRV to the Rt side of SMA, resection and re-anastomosis of the LRV in front of the SMA (transposition) was done with clamps applied to LRV without direct IVC clamping to avoid any intraoperative hemodynamic instability or potential thrombosis. Lt gonadal vein bypass to Lt common iliac vein by end to side anastomosis can drain the congested LRV and avoid dissection in the AMA and avoid also pancreatic upward retraction during the procedure which decrease postoperative risk of acute pancreatitis.

Statistical analysis:

Statistical analysis was done by SPSS v26 (IBM Inc., Chicago, IL, USA). Quantitative parametric data were presented as mean and standard deviation (SD) and were compared by paired *t*-test (or repeated measures ANOVA). Qualitative variables were presented as frequency and percentage (%) and were compared by Chi-square test or Fisher's exact test when appropriate. A two tailed *p*-value <0.05 was considered statistically significant.

Fig. (1): CT angiography reconstruction image showing acute angle of the SMA.

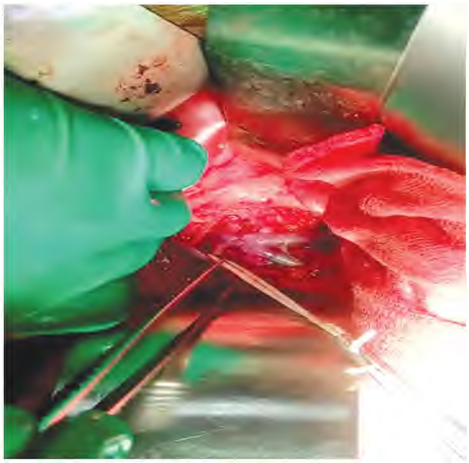

Fig. (2): Intraoperative image showing reimplantation of the LRV in front of the SMA.

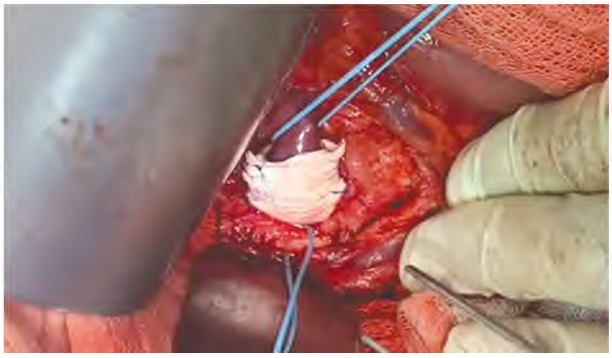

Fig. (3): External shield using PTFE ringed 8 mm double graft around the LRV.

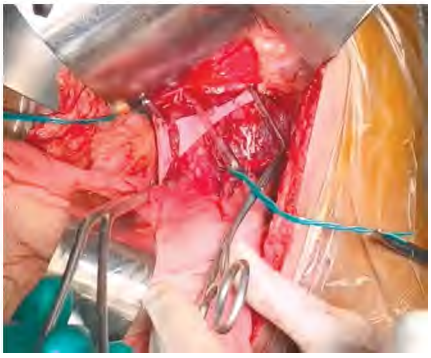

Fig. (4): Re-implantation of the LRV to the IVC few centimeters below the original compressed position.

Fig. (5): Lt gonadal vein bypass to Lt Common Iliac Vein to relief compression of LRV.

Results

The mean value (± SD) of age was 20.3 (±3.09) years. There were 8 (80%) males and 2 (20%) females. The mean value (± SD) of weight was 58.8 (±5.2) Kg. The mean value (± SD) of height was 1.7 (±0.07) m. The mean value (± SD) of BMI was 20.5 (±2.96) kg/m^2.

The mean value (± SD) of the angle was 24.3 (±4.14)°. Table (1).

Pain and hematuria decreased significantly postoperative (p<0.001 and 0.010 respectively).

Varicocele did not decrease significantly postoperative. Table (2).

The operative technique was re-implantation of LRV to IVC in 3 case (30%), transposition of LRV in front of the SMA in 4 cases, External shield using double PTFE ringed graft in one case (10%), removal of thick fibrous band from the aortomesenteric window in only one patient (10%), and Lt gonadal vein bypass to the left common iliac vein by end to side anastomosis to drain the compressed LRV without dissection at the aorto mesenteric angle. As in Table (3)

One (10%) patient had GIT symptoms. Improvement was present in 8 (80%) patients.

Complication (infection) occurred in 2 (20%) patients.

Table (1): Patient characteristics of the studied patients.

	(n=10)
Age (years)	20.3±3.09
Sex:	
Male	8 (80%)
Female	2 (20%)
Weight (kg)	58.8±5.2
Height (cm)	1.7±0.07
BMI (kg/m^2)	20.5±2.96
Angle (º)	24.3±4.14

Data are presented as mean ± SD or frequency (%).
BMI: Body mass index.

Table (2): Pain, hematuria, and varicocele of the studied patients.

	Preoperative	Postoperative	*p*-value
Pain (VAS score)	7.2±1.32	3.8±1.4	<0.001*
Hematuria	6 (60%)	0 (0%)	0..010*
Varicocele (Total 8)	6 (75%)	4 (50%)	0.170

Data are presented as mean ± SD or frequency (%).
* Significantly different as *p*-value ≤0.05.
VAS: Visual analog scale.

Table (3): Operative techniques used to decompress the LRV in the studied cases of nutcracker syndrome.

Operative technique	(n=10) (%)
Re-implanatation of LRV to IVC	3 (30%)
Transposition of LRV in front of SMA	4 (40%)
External synthetic graft shield	1 (10%)
Removal of thick bands at AMA	1 (10%)
Lt Gonadal vein to Lt common iliac bein bypass	1 (10%)

Table (4): GIT symptoms, improvement, and complication of the studied patients.

	(n=10)
GIT symptoms	1 (10%)
Improvement	8 (80%)
Complication (infection)	2 (20%)

Data are presented as frequency (%).

Discussion

This study examined the clinical and demographic characteristics of ten patients diagnosed with NCS. The findings provide valuable insights into this uncommon condition and contribute to the existing body of literature on NCS.

The mean age of patients in our study was 20.3±3.09 years. This is consistent with previous reports indicating that NCS predominantly affects young adults and adolescents **[9,10].**

However, it is noteworthy that NCS can occur across a wide age range, as cases have been reported in children as young as 4 years old **[11].**

Regarding gender distribution, our study demonstrated a male predominance, with 80% of cases being male. This finding aligns with earlier studies that have reported a higher prevalence of NCS among males **[9].** However, it has been described as occurring more frequently in females **[12].** Although a later study demonstrated equal prevalence between males and females **[6].** It is essential to acknowledge that NCS can affect individuals of any gender, and the reported gender differences may be influenced by factors such as referral patterns, symptom recognition, and healthcare-seeking behaviors. The mean BMI of patients in our study was 20.5±2.96kg/m^2, which falls within the normal range. However, low BMI might influence the development of NCS **[13].** One of the key findings of our study was the significant reduction in pain and hematuria following surgical intervention. Preoperatively, the mean pain score was 7.2±1.32, which decreased to 3.8±1.4 postoperatively.

Additionally, 60% of patients reported hematuria preoperatively, while none reported hematuria after surgery. These results align with previous studies that have demonstrated the efficacy of surgical treatment in alleviating symptoms associated with NCS, particularly pain and hematuria **[14,15].** Interestingly, our study found reduction in percentage to 50% postoperative instead of 75% preoperative without statistical difference in the presence of varicocele. This observation contrasts with some previous reports that have suggested a potential improvement or resolution of varicocele after surgical decompression of the left renal vein in NCS patients **[16-18].** However, it is essential to note that the sample size in our study was relatively small, and larger studies may be needed to further investigate the effects of surgical treatment on varicocele in NCS patients.

Our study also revealed that 10% of patients experienced GIT symptoms which is consistent with previous reports of NCS being associated with abdominal pain, nausea, and other GIT manifestations **[19].** Additionally, 80% of patients reported improvement after surgical intervention, highlighting the potential benefits of treatment for NCS patients. Notably, 20% of patients in our study experienced post-operative complications, specifically infections.

While complications are not uncommon following surgical procedures **[20]**. In comparison with other studies, our findings are largely consistent with the existing literature on NCS **[5,20-22].**

This study has several limitations that should be acknowledged. First, the sample size of ten patients is relatively small, which may limit the generaliza-

bility of the findings and the statistical power to detect significant differences. Furthermore, our study did not provide information on the follow-up duration, which could impact the observed outcomes and complicate comparisons with other studies, which could be valuable areas for future research.

Conclusions:

Our study provides valuable insights into the clinical and demographic characteristics of NCS patients, while also highlighting the potential benefits of surgical intervention in alleviating symptoms such as pain and hematuria. The findings are largely consistent with previous literature, contributing to the growing body of knowledge on this uncommon condition.

However, further research is warranted to address the limitations of our study and to explore potential risk factors, long-term outcomes, and standardized diagnostic and treatment approaches for NCS patients.

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النتائج الجراحية لمتلازمة كسارة البندق: تجربة مركز واحد

المقدمة: متلازمة كسارة البندق هي اضطراب ضغط الأوعية الدموية النادر الذي يتميز بانحباس الوريد الكلوي الأيسر ، مما يؤدي إلى مظاهر سريرية مختلفة.

الهدف من الدراسة: تهدف الدراسة الحالية إلى تقييم النتائج الجراحية للمرضى الذين يعانون من متلازمة كسارة البندق.

طرق الدراسة: تم إجراء تحليل بأثر رجعي على ١٠ مرضى مصابين بمتلازمة كسارة البندق الذين خضعوا لتدخل جراحي في مستشفيات جامعة طنطا، مصر بين يناير ٢٠٢١ وفبراير ٢٠٢٤. تنوعت التقنيات أثناء العملية الجراحية بناءً على النتائج الفردية، بما في ذلك تبديل الوريد الكلوي الايسر، ورأب الوريد التصحيحي، والتطعيم الالتفافي.

نتائج الدراسة: كان متوسط عمر المرضى 20.3 ±30.9 سنة، مع غلبة للذكور (٨٠٪). كان متوسط مؤشر كتلة الجسم 2,96±20,5 كجم/م٢، وكان متوسط الزاوية الأبهرية المساريقية درجة. بعد العملية الجراحية، لوحظ إنخفاض كبير في الألم وبيلة دموية مقارنة بمستويات ما قبل الجراحة. وأظهرت دوالي الخصية فرقاً ضئيلاً ٦٠٪ مقابل ٢٠٪. ولوحظ التحسن في ٨٠٪ من المرضى، في حين أن ٢٠٪ عانوا من إلتهابات ما بعد الجراحة.

الخلاصة: أظهر التدخل الجراحي لمتلازمة كسارة البندق نتائج واعدة، مع تخفيف كبير للألم والبيلة الدموية في المجموعة المدروسة. في حين أن تحسن دوالي الخصية كان ضئيلاً إحصائياً، فإن غالبية المرضى شهدوا تخفيفاً عاماً للأعراض.