Nutcracker syndrome complicating with renal abscess
Sevgi Yavuz,1 Aydin Ece,2 Mahmut Corapli,3 Cigdem Ilter,4 Rufat G üven5

Abstract
The nutcracker syndrome refers to compression of left renal vein between the superior mesenteric artery and aorta. Renal abscess consists of purulent and necrotic material localised to the renal parenchyma. These two entities are extremely rare and their coincidence has not previously been described in literature. Here, we report a case of a 10-year-old girl who developed left renal abscess probably due to nutcracker syndrome.

Keywords: Nutcracker syndrome, left renal vein entrapment, renal abscess, haematuria, fever.

Introduction
The nutcracker phenomenon (NCP) refers to the compression of left renal vein (LRV) most commonly between the superior mesenteric artery (SMA) and aorta. The nutcracker syndrome (NCS) is the clinical equivalent of NCP and is characterised by various symptoms such as haematuria, proteinuria, flank pain, pelvic congestion or varicocele.1,2

Renal abscess consists of purulent and necrotic material localised to the renal parenchyma.3 This is a rare entity in paediatric population and has not previously been described in NCS patients. Here we report a case of a young girl who presented with left renal abscess and was diagnosed as NCS.

Case Report
A previously healthy 10-year-old girl was admitted with fever and left flank pain lasting for six days. There was no macroscopic haematuria, trauma, urinary tract or other infections, chronic diseases, recent surgery or immunosuppressive drug usage in her medical history.

On physical examination, her weight and height were within 25-50th percentiles. The vital signs included a temperature of 38.5°C, heart rate (HR) of 90 beats/min, respiratory rate (RR) of 20 breaths/min, and blood pressure (BP) of 100/60 mmHg. Her abdomen was soft with left costovertebral angle tenderness. On urinalysis, gravity was 1015, pH 6.0, blood (+), leukocyte esterase

Figure: a) Computed tomography shows compression of left renal vein (LRV) between superior mesenteric artery (SMA) and abdominal aorta (Ao). b) 3x3 cm cystic necrotic lesion in the lower pole of the left kidney extending to peri-renal space.

1-3Division of Pediatric Nephrology, 4Department of Radiology, 5Department of Pediatrics, Dicle University, Diyarbakir, Turkey.

Correspondence: Sevgi Yavuz. Email: drsyavuz@gmail.com
(+), protein (-), and urine microscopy showed five to ten red and white blood cells (RBCs and WBCs). Serum biochemistry, haemoglobin and platelet counts were within normal ranges. WBC count (21000/mm³), sedimentation rate (40 mm/h) and serum C-reactive protein (CRP) (65 mg/L) levels were increased. Abdominal ultrasound (US) showed a single iso-hypoechoic lesion on left kidney. Colour Doppler US revealed centrally necrotic and vascular hyperheterogenous solid lesion in the lower pole of the left kidney with enlargement of LRV proximal to the aorto-mesenteric junction. Computed tomography (CT) demonstrated the compression of LRV between SMA and abdominal aorta (Ao) (Figure-a) and a 3x3cm cystic necrotic lesion in the lower pole of the left kidney extending to peri-renal space (Figure-b). A diagnosis of abdominal abscess with NCS was established and vancomycin-meropenem combination therapy was empirically started. Percutaneous aspiration was not performed because of disapproval of her parents. The initial urine and blood culture samples were sterile, and voiding cystourethrogram (VCUG) imaging was normal. The antibiotic therapy continued for four weeks and the patient was closely followed-up with serial US. At the end of the treatment, clinical and US findings became normal whereas microscopic haematuria persisted for the next six months.

**Discussion**

The LRV entrapment of SMA and aorta was first described in 1950. In 1972, the disorder was referred to as NCS. It is very rare and usually diagnosed late because of the variability of symptoms and absence of consensus on diagnostic criteria. Many patients, especially children, are asymptomatic. The most common clinical feature, haematuria is attributed to high LRV pressure resulting in the rupture of the thin-walled septum between the small veins and the collecting system in renal fornix. It varies from micro- to macro-haematuria, depending on the severity of LRV hypertension. The present case remained asymptomatic until a pyogenic kidney lesion occurred. Despite the full recovery of abscess, microscopic haematuria persisted during the follow-up. Furthermore, she did not suffer from proteinuria, fatigue, pain or pelvic congestion which constitute the common findings of NCS.

Renal abscess is an uncommon, but severe form of renal infection in children. Patients may present with fever, chills, abdominal or flank pain, weight loss and malaise. Three basic pathophysiological mechanisms have been described for the development of renal abscess. 1) Haematogenous spread, particularly in cases with sterile urine culture. These lesions are primarily located in the renal cortex which has abundant blood vessels and extensive lymph drainage. 2) Ascending infections due to reflux or stasis of infected urine which predominantly locates on cortico-medullary area. 3) Direct spread of a contiguous infection.

In our patient, renal cortical location, lack of evidence of urinary tract or a neighbouring infection indicated a haematogenous spread. Additionally, normal VCUG excluded the possibility of vesicoureteral reflux which mainly predisposes to ascending infections.

The predominant causative agents of renal abscess are S. aureus and E. Coli. Unfortunately, a pathogen was not identified in our patient because of disapproval for aspiration from the lesion. Most abscesses are unilateral and occur in the right kidney. Contrarily, our patient had left-sided renal abscess. Ipsilateral localisation of abscess with LRV entrapment brought to mind a question whether these two conditions were related or not. However, the coexistence of renal pyogenic lesions with NCS has not previously been reported in literature. On the other hand, the blood flow within the abscess suggested that rupture of renal microvarices might have promoted the development of abscess in our case with NCS. The lack of a predisposing situation like immune-suppression or a known origin of infection supported our opinion.

Ultrasonography and CT scanning are the most common radiological tools for determining the abscess. Both modalities reveal a hypoechoic or hypodense mass. The real-time Doppler US is recommended as the first diagnostic test in patients with suspected NCS. CT scan and CT angiography are accepted as confirmatory imaging techniques.

The initial treatment of renal abscess consists of prolonged course of broad-spectrum antibiotics. For renal abscess >5cm or those that fail to respond to antibiotic therapy, percutaneous drainage should be considered. On the other hand, the treatment of NCS is controversial. Conservative approach is more acceptable for mild cases and also for pubertal children because of high rate of spontaneous resolution probably due to physical development. Endovascular or open surgery might be recommended in severe conditions.

**Conclusion**

The present case is the first demonstrating coincidence of NCS with renal abscess in literature. The rupture of microvarices in NCS may predispose to a pyogenic kidney infection. Renal abscess should be considered in patients...
with unexplained fever of origin and flank pain.

References