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# **Case Report**

# Nutcracker syndrome unveiled by severe Typhoid fever: A rare case report\*

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#### ABSTRACT

Typhoid fever, caused by Salmonella Typhi, is a severe bacterial infection prevalent in developing countries, and can result in life-threatening complications if untreated. Nutcracker Syndrome is a rare vascular disorder involving compression of the left renal vein between the aorta and the superior mesenteric artery. It can lead to various symptoms and poses diagnostic and management challenges. We present a case study of a patient diagnosed with typhoid fever in a Unit of Critical Emergency Care. Coincidentally, the evaluation through CT-scan revealed the presence of Nutcracker Syndrome. This report underscores the incidental discovery of Nutcracker Syndrome during the assessment of a patient with typhoid fever in a critical emergency care setting.

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# Introduction

Typhoid fever, caused by the bacterium Salmonella enterica serovar Typhi, continues to be a significant global health concern, particularly in regions with inadequate sanitation and limited access to clean water.

Severe typhoid fever, characterized by a constellation of debilitating symptoms including high fever, gastrointestinal distress, and systemic complications, can result in lifethreatening scenarios requiring critical care support. The admission of typhoid fever patients to the intensive care unit (ICU) underscores the complex and evolving nature of this infectious disease, with potentially grave outcomes that demand a multidisciplinary approach.

Nutcracker Syndrome is an uncommon vascular disorder characterized by the compression of the left renal vein between the aorta and the superior mesenteric artery. This anatomical anomaly can result in a variety of clinical manifestations, including flank pain, hematuria, and pelvic congestion. The complex interplay of anatomical and hemodynamic factors contributes to the diagnostic complexity

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and necessitates tailored management approaches for this condition.

We report in this observation, a severe typhoid fever in a patient in Critical Emergency Care. Incidentally, CT evaluation unveiled Nutcracker Syndrome. This highlights the chance identification of Nutcracker Syndrome during typhoid fever assessment in emergency care.

## Patient and observation

### Patient information

A 37-year-old male with no significant past medical history presented with a constellation of symptoms, including fever, diarrhea, and vomiting. The patient reported a substantial weight loss of 10 kilograms over a relatively short period. These symptoms collectively raised concerns regarding a potential underlying medical condition.

#### Clinical findings

Upon initial assessment, the patient exhibited persistent fever, with recorded temperatures exceeding the normal range. The presence of fever, along with gastrointestinal symptoms such as diarrhea and vomiting, prompted an evaluation for infectious etiologies. Notably, the patient's weight loss was a prominent feature, underscoring the severity and chronicity of the presenting illness.

Upon detailed clinical examination, the patient presented with a visibly weakened and emaciated appearance. Physical assessment revealed signs of dehydration, including dry mucous membranes and reduced skin turgor. The patient appeared fatigued and lethargic, with notable muscle wasting evident in the extremities. The patient's weight is recorded as 51 kilograms. Height is measured at 185 centimeters.

The calculated body mass index based on the provided weight and height is approximately 14.9.

Vital signs indicated an elevated heart rate of 140 bpm and increased respiratory rate of 35 c/min, both of which were consistent with the patient's fever and potential compensatory responses to metabolic disturbances. Blood pressure readings remained within acceptable ranges albeit on the lower end at 95/75 mmHg, underscoring the importance of monitoring for potential hemodynamic instability.

Abdominal examination revealed diffuse tenderness on palpation, particularly in the right lower quadrant. Bowel sounds were diminished, indicative of potential gastrointestinal motility disruptions.

Urinalysis indicated signs of concentrated urine, in line with the observed dehydration. Electrolyte imbalances, including hyponatremia at 127mmol/L (135-145 mmol/L) and severe hypokalemia at 1,9 mmol/L (3.5-5 mmol/L), were evident, further highlighting the need for meticulous electrolyte monitoring and correction.

The arterial blood gas analysis revealed severe alkalosis, characterized by a pH of 7.62 (7.38-7.42), an elevated HCO3level of 59 mmol/L (22-26 mmol/L), and a PCO2 reading of 64 mmHg (35-45 mmHg). Collectively, the clinical examination provided valuable insights into the patient's deteriorating physiological state, suggesting a severe systemic illness with significant involvement of multiple organ systems. The combination of physical findings, laboratory abnormalities, and the patient's reported symptoms guided the subsequent diagnostic approach and therapeutic interventions in the intensive care unit.

#### Microbiological investigations

Blood cultures obtained during the initial assessment revealed the presence of Salmonella enterica serovar Typhi, confirming the active infection. The isolation and identification of the pathogen from blood samples provided direct evidence of the systemic dissemination of the causative agent, further validating the clinical suspicion.

Serology Widal: The positive Widal serology results provided essential indirect evidence of recent or ongoing infection with S.Typhi. The elevated titers of specific antibodies against O and H antigens were consistent with the clinical presentation and microbiological findings. The combination of positive serology, along with microbiological confirmation, reinforced the diagnosis of typhoid fever.

Diagnostic Imaging: An angioscan (CT) of the patient's abdomen and pelvis revealed a pronounced compression of the left renal vein between the aorta and the superior mesenteric artery, consistent with Nutcracker Syndrome (Fig. 1), and allowed us to eliminate a mesenteric ischemia diagnosis. This anatomical anomaly led to a significant narrowing of the left renal vein lumen, causing venous congestion in the left kidney. The right renal vein appeared normal. The findings highlight the presence of Nutcracker Syndrome as an incidental finding in the context of the patient's typhoid fever diagnosis.

#### Therapeutic interventions

The confirmed diagnosis of typhoid fever paved the way for the implementation of a targeted and tailored treatment strategy, encompassing antimicrobial therapy, intravenous fluid resuscitation, electrolyte correction such as potassium refill, and meticulous supportive care.

Close monitoring of renal function and hemodynamic status is imperative due to potential exacerbation of the symptoms.

Prompt hemodynamic recovery and symptom resolution obviated the need for endovascular or surgical intervention to address Nutcracker Syndrome.

#### Follow-up and outcome of interventions

Following the initiation of targeted antibiotic therapy for typhoid fever and concurrent management of Nutcracker Syndrome-associated symptoms, the patient exhibited notable clinical improvement. Comprehensive fluid resuscitation and electrolyte correction were instrumental in restoring the patient's hydration and overall well-being.

Serial monitoring of renal function and hemodynamic parameters showed favorable trends. The patient's symptoms gradually abated.



Fig. 1 – CT scan showing compression of the left renal vein (LRV) between the aorta and the superior mesenteric artery, consistent with Nutcracker Syndrome.

# Discussion

Severe typhoid fever, caused by Salmonella Typhi, continues to be a significant global health concern, particularly in resourcelimited regions. Patients with severe typhoid fever often require ICU management due to life-threatening complications [1].

Studies have highlighted the diverse spectrum of severe manifestations, including septic shock, multiorgan dysfunction, and encephalopathy [2,3]. The ICU plays a pivotal role in providing aggressive fluid resuscitation, antibiotic therapy, and hemodynamic support [4].

Nutcracker Syndrome (NCS) is a rare vascular disorder characterized by compression of the left renal vein between the aorta and the superior mesenteric artery, resulting in various clinical manifestations. NCS was first described in the medical literature by El-Sadr and Mina in 19501. Since then, numerous studies have contributed to a deeper understanding of its clinical presentation, diagnostic methods, and management strategies [5].

Patients with NCS may present with a diverse range of symptoms, including hematuria, abdominal pain, flank pain, pelvic congestion syndrome, and even varicocele in males. The clinical spectrum and severity of symptoms can vary widely among patients, leading to challenges in diagnosis [6].

Diagnosis of NCS often involves a combination of clinical evaluation and imaging studies. Noninvasive imaging modalities such as Doppler ultrasonography, computed tomography angiography, and magnetic resonance angiography are commonly employed to visualize the anatomic relationship between the vessels and assess venous flow dynamics. In our case, imaging played a crucial role in discovering the Nutcracker Syndrome, without it the vascular disorder might have gone unnoticed as it's symptoms can overlap with those of typhoid fever.

One of the major challenges in diagnosing NCS is differentiating it from other conditions that may present with similar symptoms, such as renal stones, urinary tract infections, or pelvic congestion syndrome. This highlights the importance of comprehensive clinical evaluation and appropriate imaging techniques [7].

The occurrence of typhoid fever in conjunction with Nutcracker Syndrome is exceptionally uncommon with no similar cases reported in the literature.

In cases of severe typhoid fever associated with Nutcracker Syndrome, a multidisciplinary approach is paramount. Antibiotic therapy targeting the Salmonella Typhi infection is initiated promptly. Close monitoring of renal function and hemodynamic status is imperative due to potential exacerbation of Nutcracker Syndrome-related symptoms. Fluid resuscitation and electrolyte balance maintenance are crucial.

Invasive interventions, such as endovascular stenting to alleviate left renal vein compression, may be considered if hemodynamic compromise persists [8]. Surgical options, including renal vein transposition, could be explored if conservative measures prove inadequate [9]. A comprehensive assessment of the patient's clinical status guides the selection of optimal therapeutic strategies [10].

This case underscores the significance of timely intervention, multidisciplinary care, and tailored therapeutic strategies in achieving positive outcomes for patients with combined typhoid fever and Nutcracker Syndrome.

## Conclusion

In conclusion, the co-occurrence of typhoid fever and nutcracker syndrome is an exceedingly rare phenomenon. The intricate interplay between these distinct conditions presents diagnostic challenges and necessitates a multidisciplinary approach for effective management. As medical literature on this association remains limited, heightened clinical awareness is essential to timely recognize and address this unique combination of pathologies. Further research and reporting of such cases are imperative to enhance our understanding of this uncommon clinical intersection and to guide optimal patient care.

#### What is already known about it

- In cases of severe typhoid fever associated with Nutcracker Syndrome, a multidisciplinary approach is paramount.
- The co-occurrence of typhoid fever and nutcracker syndrome has been very rarely described in the literature,

#### What this study provides

- The interest of presenting this case lies in the exceptional occurrence of nutcracker syndrome being revealed by typhoid fever.
- By presenting this case, we discuss diverse therapeutic interventions applicable to both typhoid fever and nutcracker syndrome.

## **Patient consent**

Informed consent was approved by the patient being fully aware of the stakes of this case report and wanting willingly to be a part of it.

#### REFERENCES

- [1] Date KA, Newton AE, Medalla F, Blackstock A, Richardson LC, McCullough A, et al. Changing patterns in enteric fever incidence and increasing antibiotic resistance of enteric fever isolates in the United States, 2008–2012. Clin Infect Dis 2015;61(4):449–57.
- [2] Parry CM, Hien TT, Dougan G, White NJ, Farrar JJ. Typhoid fever. N Engl J Med 2002;347(22):1770–82.
- [3] Wain J, Diep TS, Bay PV, Walsh AL, Vinh H, Duong NM, et al. Specimens for the diagnosis of typhoid fever: analysis of 197 cases. Clin Infect Dis 1998;27(4):739–46.
- [4] Arora A, Tyagi P. Intensive care management of typhoid fever in children. J Trop Pediatr 2016;62(1):1–9.
- [5] El-Sadr AR, Mina E. The "nutcracker" action of the renal vein: a cause of ureteral obstruction. J Urol 1950;63(2):203–6.
- [6] Shin JI, Park JM, Lee JS, Kim MJ. Effect of renal Doppler ultrasound on the detection of nutcracker syndrome in children with hematuria. Eur J Pediatr 2007;166(5):399–404.
- [7] Kurklinsky AK, Rooke TW. Nutcracker phenomenon and nutcracker syndrome. Mayo Clin Proc 2010;85(6):552–9.
- [8] Eskandari MK, Shandera KC, Ignatoff JM, Flinn WR. Open and endovascular treatment of symptomatic "nutcracker" phenomenon. J Vasc Surg 2002;36(5):853–8.
- [9] Zhang H, Zhang J, Zhang Z, Chen L, Wang H. Nutcracker syndrome treated with left renal vein transposition in two pediatric patients. J Pediatr Surg 2014;49(1):147–50.
- [10] Kurklinsky AK, Rooke TW. Nutcracker phenomenon and nutcracker syndrome. Mayo Clinic Proc 2010;85(6):552–9.