

Left Renal Vein Thrombus and Pulmonary Thromboembolism Caused by Nutcracker Syndrome: a Case Report

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Case report

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Abstract

Background: Nutcracker syndrome (NCS) refers to compression of the left renal vein (LRV) between the aorta and superior mesenteric artery (SMA), which results in renal venous hypertension and its resultant clinical manifestations. Left renal vein thrombus (LRVT) complicating NCS is relatively rare. To the best of our knowledge, there are only four case reports of LRVT complicating NCS. Furthermore, there are no reports of pulmonary thromboembolism (PTE) caused by NCS. Herein, we describe a rare case of NCS causing LRVT and PTE and its clinical management.

Case Presentation: A 40-year-old man was admitted to our hospital with acute left flank pain. Computed tomography angiography (CTA) revealed compression of the LRV between the aorta and the SMA with an LRVT. Furthermore, CTA revealed bilateral PTE. Rivaroxaban was administered as an anticoagulant. Twenty days after initiation, CTA revealed complete resolution of PTE and LRVT, and repeat CTA at 3 and 6 months showed no recurrence.

Conclusions: This case report demonstrates that NCS may be a possible cause of LRVT and PTE. We review the reported cases of NCS complicated by LRVT and discuss the imaging modalities for NCS.

Background

Nutcracker syndrome (NCS) refers to compression of the left renal vein (LRV) between the aorta and superior mesenteric artery (SMA), which results in renal venous hypertension and its clinical manifestations [1]. The characteristic clinical features of NCS include hematuria, abdominal pain, and left gonadal vein varices. Left renal vein thrombus (LRVT) complicating NCS is relatively rare. To the best of our knowledge, there are only four case reports of LRVT complicating NCS [2–5].

Pulmonary thromboembolism (PTE) is a potentially fatal disease. The overall mortality rate of PTE ranges from 8.1% in stable cases to 25% in patients presenting with cardiogenic shock and 65% in patients requiring cardiopulmonary resuscitation [6]. PTE refers to the obstruction of the pulmonary vessels by a thrombus, and the majority (> 90%) of cases of PTE is caused by deep vein thrombosis in the pelvis, thigh, or lower leg. To the best of our knowledge, there are no reports of PTE caused by NCS. Herein, we describe the case of a 40-year-old man presenting with asymptomatic LRVT and PTE complicating NCS. We report this case and discuss its clinical features, and summarize the available literature of LRVT associated with NCS.

Case Presentation

A 40-year-old man was admitted to our clinic with acute left flank pain. His past medical history included a right ureteral stone and intestinal injury due to a car accident. His vitals were stable, and examination revealed isolated left costovertebral angle tenderness. Physical examination revealed a height of 182 cm and a weight of 73 kg with a body mass index of 21.8 kg/m². He was not taking any regular medications. Regarding his social history, he was a truck driver. Urinalysis revealed microscopic hematuria (5–9/high

powered field). The patient's laboratory data revealed impaired renal function (serum creatinine, 1.13 mg/dL) and raised D-dimer levels (4.9 µg/mL). Doppler ultrasonography (USG) revealed compression of the LRV between the abdominal aorta and SMA, as well as an LRVT.

Computed tomography angiography (CTA) was performed for better visualization. This revealed compression of the LRV between the aorta and the SMA with an LRVT, and the distance between the Ao and SMA at the level of the LRV was 2.5 mm (Fig. 1). Moreover, sagittal CTA showed that the angle between the Ao and SMA was 22° (Fig. 2). We also measured the peak velocity (PV) at the hilar portion of the LRV and at the LRV between the aorta and SMA using USG. The PV was 18.7 cm/s and 107 cm/s, respectively, and the ratio of PV in the LRV between the aorta-SMA portion and the hilar portion was 5.7. The pretreatment CTA revealed that the LRVT was found distal to the compression of the LRV between the Ao and SMA. Based on the above findings, the patient was diagnosed with NCS, and this was suspected to be the cause of the LRVT. Furthermore, CTA revealed bilateral PTE (Fig. 3). There were no findings of deep vein thrombosis or thrombosis other than the LRVT on USG and CTA. Therefore, the pathogenesis of PTE appears to be the LRVT. Laboratory data and imaging studies excluded the presence of other thrombogenic factors, such as malignant neoplasm, vascular malformation, trauma, heritable thrombophilia, protein C/S deficiency, and antiphospholipid syndrome. Thus, his only thrombogenic risk factor was prolonged sitting due to his job as a truck driver. Based on the above, he was diagnosed with PTE caused by LRVT.

The patient was reviewed by the cardiology team, and he was started on rivaroxaban 30 mg daily for anticoagulation. Since the PTE was mild, the cardiologists concluded that an inferior vena cava filter was unnecessary. Ten days after the initiation of rivaroxaban, a blood test revealed improvement in renal function (serum creatinine was 0.8 mg/dL) and normalization of D-dimer (1.0 µg/mL), and CTA revealed the resolution of the PTE and a decrease in the size of the LRVT (Fig. 4). Twenty days after initiation, CTA revealed complete disappearance of the PTE and LRVT (Fig. 5). Thereafter, rivaroxaban was decreased to 20 mg daily. USG and CTA, repeated 3 and 6 months later, showed no recurrence of PTE and LRVT. After carefully reviewing the patient's history and risk factors, no other causes of LRVT after PTE were identified. And from now, there were no adverse event from the induction of rivaroxaban.

Discussion And Conclusions

NCS was first described by De Schepper in 1972 [7]. NCS refers to the compression of the LRV between the aorta and SMA, which results in renal venous hypertension and its clinical features. The most common clinical manifestations of NCS include hematuria, abdominal pain, and left gonadal vein varices. LRVT as a complication of NCS is relatively rare. To the best of our knowledge, there are only four case reports of LRVT complicating NCS [2–5].

Furthermore, there are no reports of PTE caused by NCS. Nakashima et al. reviewed four cases of LRVT associated with NCS [5]. Three of the four cases of LRVT presented with left flank pain and showed macroscopic and/or microscopic hematuria, similar to our case. Membranous glomerulonephritis, use of

oral contraceptives, and endometrial cancer were identified as risk factors for a hypercoagulable state. In our case, the patient's job as a truck driver was the only thrombogenic factor. PTE did not occur in any of the four aforementioned patients. All four patients received anticoagulation therapy (warfarin only or heparin followed by warfarin).

The diagnosis of NCS requires relatively invasive examinations such as venographic imaging or intra-arterial digital subtraction angiography. Less invasive examinations such as ultrasonography and CTA can be used as alternative modalities for the diagnosis of NCS. Kim et al. used USG and measured the PV at the hilar portion of the LRV and at the LRV between the aorta and the SMA [8]. They found that the cutoff value that may be useful for the diagnosis of NCS is a PV ratio of more than 5.0. Fu et al. used CTA and measured the angle between the aorta and the SMA [9]. The angles were $39.3 \pm 4.3^\circ$ in the NCS group and $90 \pm 10^\circ$ in the control group. They also measured the distance between the SMA and aorta at the level of the left renal vein. The distances were 12 ± 1.8 mm in the control group and 3.1 ± 0.2 mm in the four patients, respectively. Our case met all these criteria; therefore, the patient was diagnosed with NCS.

This report demonstrates that NCS is a possible cause of LRVT and PTE.

List Of Abbreviations

NCS, Nutcracker syndrome

LRVT, left renal vein thrombus

CTA, computed tomography angiography

USG, ultrasonography

SMA, superior mesenteric artery

PTE, pulmonary thromboembolism

PV, peak velocity

Declarations

Ethics approval and consent for participate

Informed consent for participate was obtained from the patient.

There are the written consent in Japanese and copy of the consent is available for review.

Ethical approval was granted by the Otaru General Hospital.

Consent for publication

Informed consent for the publication was obtained from the patient.

Availability of data and materials

The datasets used and analyzed during the present study are available from the corresponding author upon reasonable request.

Competing interest

There are no financial or competing interests to declare.

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Authors' contributions

KH, SY, and MK designed the study, collected, and analyzed the clinical data. KH wrote the manuscript. NY and YS contributed to the conception and design of the study and revised the manuscript. All authors have read and approve the manuscript.

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Figures



Figure 1

Contrast-enhanced computed tomography angiography reveals compression of the left renal vein between the aorta (Ao) and the superior mesenteric artery (SMA) with a left renal vein thrombus (yellow arrow) and the distance between the Ao and SMA at the level of the left renal vein is 2.5 mm.

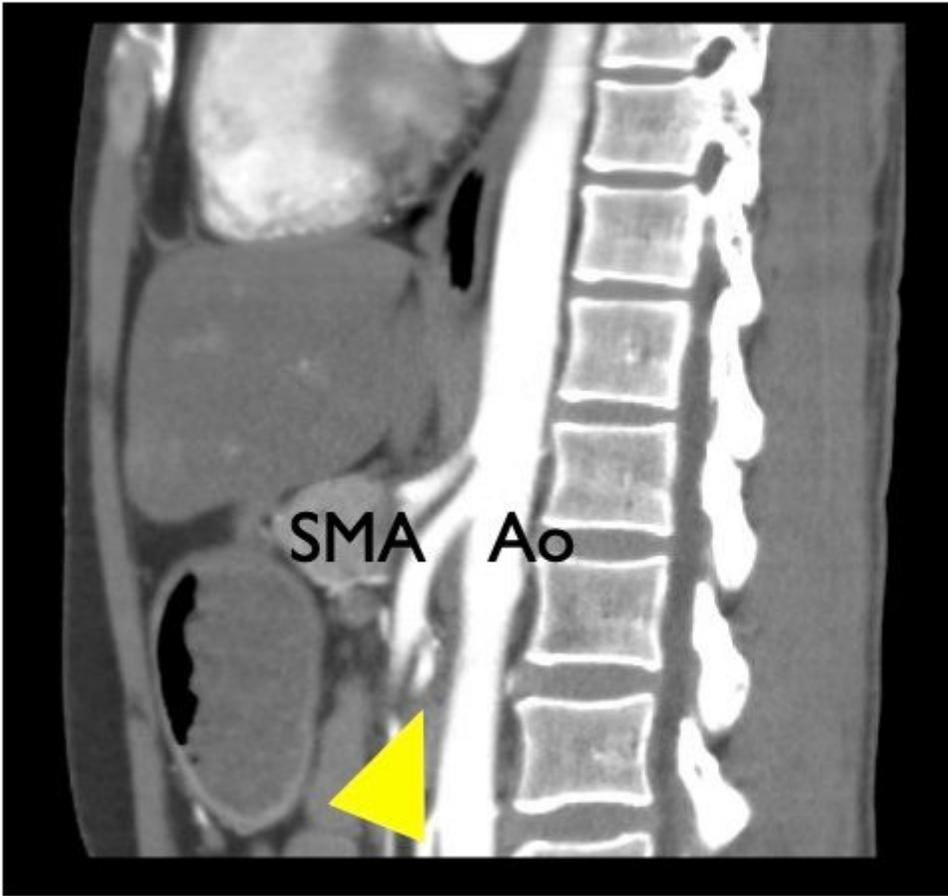


Figure 2

The sagittal computed tomography angiography shows that the angle between the aorta and superior mesenteric artery is 22°.



Figure 3

The computed tomography angiography also reveals bilateral pulmonary thromboembolism.

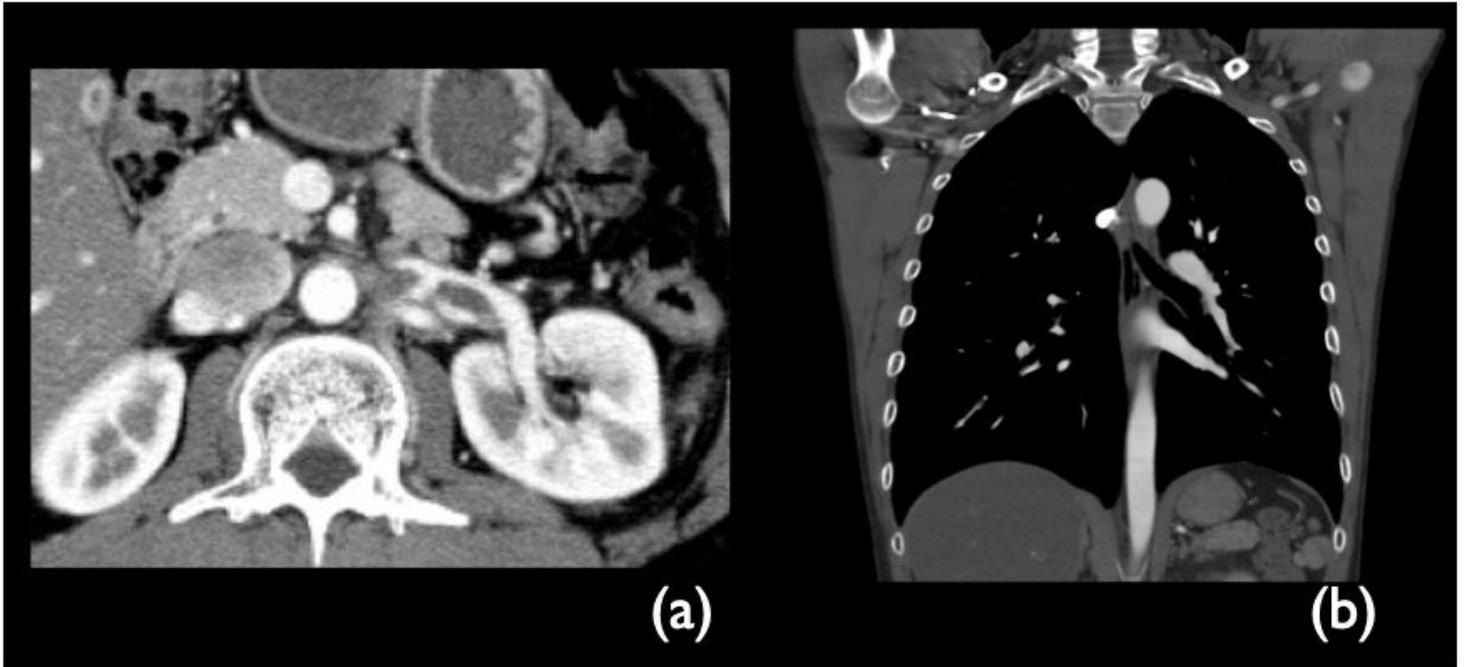


Figure 4

Ten days after the initiation of rivaroxaban, the computed tomography angiography reveals a reduction in the size of the left renal vein thrombus (a) and the disappearance of the pulmonary thromboembolism(b).

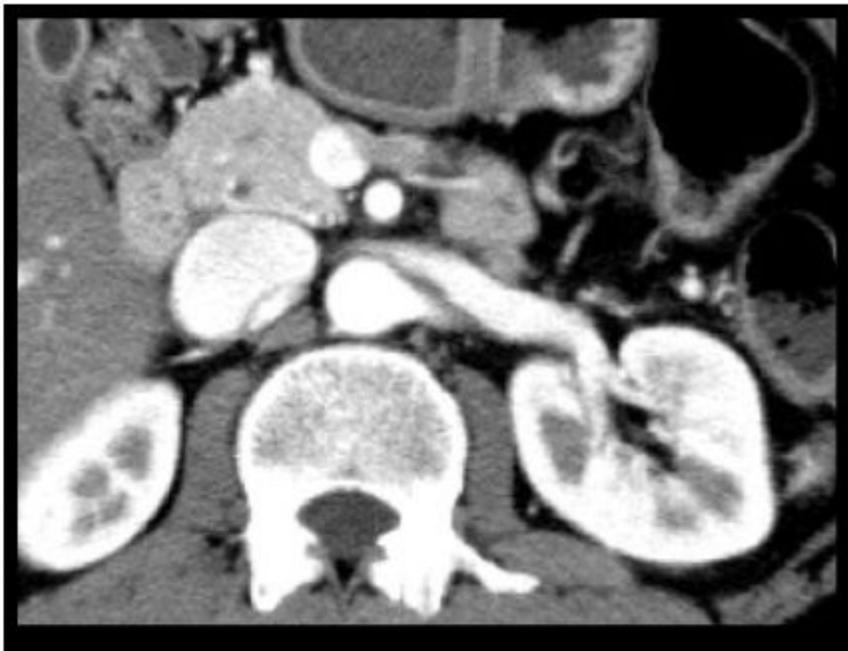


Figure 5

Twenty days after initiation of rivaroxaban, the computed tomography angiography reveals the complete disappearance of the left renal vein thrombus.

Supplementary Files

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- [CAREChecklistofPTEcausedbyNCS.docx](#)
- [CAREchecklistKantahori.pdf](#)