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Nutcracker Syndrome- compressed literature review

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ABSTRACT

The Nutcracker Syndrome (NCS) is a rare and ununderstood condition. The leading cause is the compression of the left renal vein (LRV), which can lead to symptoms such as hematuria, left flank pain, or proteinuria. Other rare symptoms, such as hypertension and headaches, have also been reported. There are cases where individuals exhibit radiological features of LRV compression without clinical symptoms, referred to as the Nutcracker Phenomenon (NCP). Additionally, NCS can be associated with other disorders such as varicocele, pelvic congestion syndrome, superior mesenteric artery syndrome, or loin pain hematuria syndrome. Based on the compression location, it can be classified into anterior and posterior types. Due to its rarity and discrepancies in radiological criteria, diagnosis can be challenging. However, there are currently many effective and safe treatment methods available. In this paper, we have attempted to review the available literature to gather, condense, and present the current knowledge on this topic.

Keywords: "Nutcracker Syndrome (NCS)", "Left Renal Vein Entrapment", "Treatment", "Diagnostic", "Literature Review".

1. INTRODUCTION

The Nutcracker Syndrome (NCS), also known as Left Renal Vein Entrapment Syndrome, can be described as a constellation of clinical symptoms associated with the obstruction of venous blood outflow from the kidney. The exact mechanism is not fully understood, but existing theories indicate the role of venous congestion in developing the disease (Kolber et al., 2021). Congestion is most commonly caused by the external compression of the left renal vein (LRV) between the abdominal aorta and the superior mesenteric artery (SMA) (Granata et al., 2021; He et al., 2014). When not accompanied by clinical symptoms, this finding is referred to as the Nutcracker Phenomenon (NCP) (He et al., 2014; Kurklinsky and Rooke, 2010).

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It should also be noted that NCS can occur during other diseases or anatomical anomalies or coexist with other conditions, which may present diagnostic challenges. Moreover, although it is classified as a rare disease, its prevalence may actually be more widespread (Yoon et al., 2022). This study aimed to present the currently available knowledge about NCS, highlight the difficulties associated with diagnosis, compare diagnostic and therapeutic algorithms, and emphasize the still unknown aspects of this syndrome.

2. METHODOLOGY

Search Strategy

To gather and catalog relevant articles, we searched the "PubMed" database for literature related to NCS from 2000 to May 2024. The keywords used included "Nutcracker syndrome", "NCS", "Nutcracker Syndrome Case Report", "Nutcracker Syndrome Review", and "Left Vein Entrapment Syndrome".

Inclusion and Exclusion Criteria

The studies needed to be related to Nutcracker Syndrome and address issues such as symptoms, classification, development, epidemiology, and especially diagnostics and treatment. Additionally, particular attention was given to aspects not previously described in other literature review publications. The studies had to be published in peer-reviewed journals and written in English. Studies that did not meet the criteria mentioned above were excluded.

Data Extraction

Selected articles were then read and analyzed, focusing on new information and discoveries related to NCS. Special effort was made to search for and analyze individual diagnostic algorithms and discrepancies in the proposed diagnostic criteria.

Data Synthesis

A descriptive analysis was performed. The information has been further organized into Introduction, Classification, Symptoms and Complications, Epidemiology, Diagnosis, Treatment, Discussion, and Conclusion. The information presented in each section has been condensed to provide a comprehensive overview of the available knowledge regarding NCS.

3. RESULT AND DISCUSSION

Classification

The classic Nutcracker Syndrome can be divided into anterior and posterior types. The anterior type is understood as the compression of the left renal vein between the superior mesenteric artery and the aorta (Orczyk et al., 2017) (Figure 1, Diagram 1). The compression of LRV is complex and multifactorial, involving findings such as a reduced aortomesenteric angle and reduced distance between the aorta and the superior mesenteric artery at the passage of the LRV. Other factors include an asthenic body build and ptosis of the left kidney (Reed et al., 2009; Kim et al., 2011). Additionally, the visceral fat measurements seem to correlate with the value of the aortomesenteric angle, and it is postulated that an increase in its amount causes an increase in the angle, which may be one of the components of the spontaneous remission of this syndrome.

The relationship between the amount of adipose tissue and NCS is unclear. It appears that in adults, subcutaneous fat correlates better with BMI than visceral fat. It appears that in adults, subcutaneous fat correlates better with BMI than visceral fat. Additionally, evidence suggests that the amount of visceral adipose tissue may correlate more strongly with the SMA angle, LRV distance, and the distance from the duodenum in women than in men. There are clinical indications that acquiring more body fat, hence higher BMI, led to a greater amount of visceral fat tissue, resulting in frontal displacement of SMA and, therefore, spontaneous resolution of NCS. BMI also correlates with improving the hemodynamics of the kidneys (Shin et al., 2005; Arthurs et al., 2012). The degree of renal vein compression correlates with the presence and severity of clinical symptoms (Hangge et al., 2018).

The posterior Nutcracker Syndrome is rarer and is associated with the retroaortic or circumaortic course of the left renal vein. (Figure 1, Diagrams 2 and 3). This course causes the compression of the vein by the aorta and the spine (Park et al., 2018; Shi et al., 2018). During embryonic development, numerous periaortic anastomoses, known as the aortic collar, participate in the formation of the left renal vein. Due to the atrophy of the anterior part or incomplete atrophy of the posterior part of these connections, an atypical vascular connection may develop, forming the basis of the posterior nutcracker syndrome (Orczyk et al., 2017; Farina et al., 2023). Shi et al., (2018) noted in their studies that the circumaortic type is rarer and less likely to cause symptoms than the type with a single retroaortic vessel.

Other causes of renal vein compression manifesting as NCS have also been described in the literature. These causes include aortic aneurysms Chansakaow and Orrapin, (2023), aortic dissection Kodama et al., (2013), duplication of the inferior vena cava Midenberg et al., (2021), vascular anomalies associated with the celiac trunk Arthurs et al., (2012), lymphadenopathy Tahara et al., (2022), scoliosis Kolber et al., (2021), rapid weight loss due to bariatric surgery Camacho-Fernández-Pacheco et al., (2017), and pregnancy (Itoh et al., 1997). Right-sided Nutcracker Syndrome is extremely rare. There are only a few case reports in the literature that were associated with factors such as a left-sided inferior vena cava or pregnancy (Yildiz et al., 2014; Radisic et al., 2007).

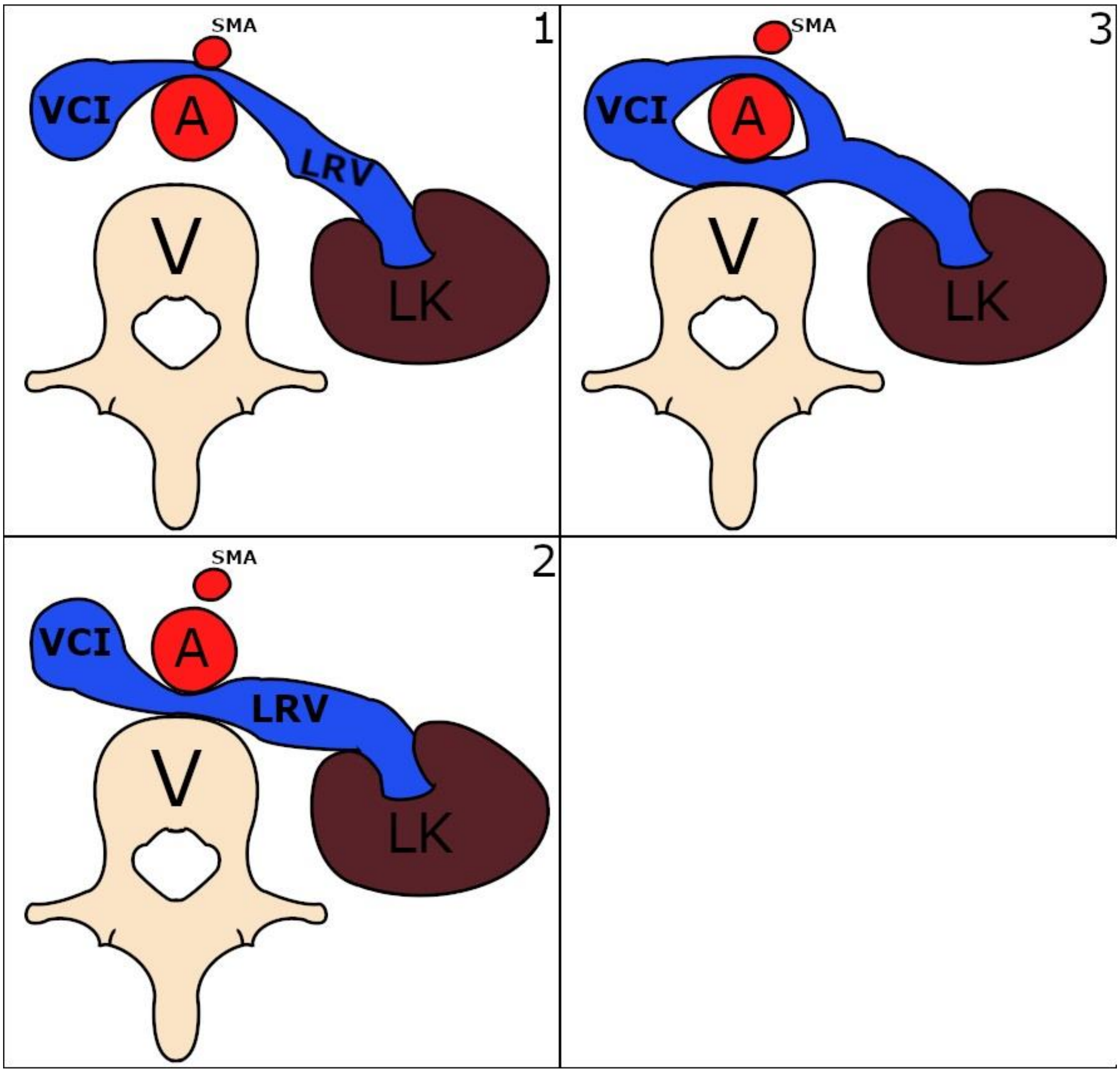

Figure 1 Schematic representation of different vascular variants occurring in "Nutcracker Syndrome". The structures are labeled as follows: "A"-Aorta, "LK"-Left Kidney, "LRV"-Left Renal Vein, "SMA" – Superior Mesenteric Artery, "V"-Vertebrae, "IVC"-Inferior Vena Cava. Diagram 1 illustrates "Anterior Nutcracker Syndrome", where the "LRV" is compressed between the aorta and the "SMA". Diagram 2 illustrates "Posterior Nutcracker Syndrome", where the "LRV" is compressed between the aorta and the spine. Diagram 3 also illustrates "Posterior Nutcracker Syndrome" with a vascular variant where, in addition to the preaortic course of the "LRV", there is an additional retroaortic branch.

Symptoms and Complications

Symptoms associated with NCS include hematuria, left flank pain (being referred to as the most frequent symptoms), proteinuria, and varicocele (Orczyk et al., 2016; Nastasi et al., 2022). In cases of hematuria, the severity ranges from microhematuria to significant bleeding, even causing anemia (Vianello et al., 2016). Prolonged compression of the left renal vein, along with increased vascular pressure, can lead to the development of collateral circulation through the left gonadal vein, ureteral veins, and the second or third communicating lumbar veins (Kurklinsky and Rooke, 2010; Ananthan et al., 2017). Left flank pain can be caused by congestion of the vertebral plexus resulting from increased pressure in the gonadal vein (Berthelot et al., 2022). Increased blood flow through the gonadal vein results in its dilation, which can lead to left-sided varicocele in men (even in children) and pelvic congestion syndrome (PCS) in women (Kurklinsky and Rooke, 2010; Reddy and Shekar, 2020).

Varicocele, in turn, manifests as scrotal pain, abnormalities in semen parameters, or differences in testicular size (Reddy and Shekar, 2020; Li et al., 2018). Additionally, men suffering from NCS with varicocele have a higher chance of recurrence of varicocele after surgical variocelectomy (Li et al., 2018). PCS is a chronic pelvic pain syndrome lasting more than six months, which can be accompanied by additional symptoms, such as lower back pain and a feeling of heaviness in the pelvis, which can be exacerbated by factors such as menstrual bleeding or sexual intercourse (Xie et al., 2023; Basile et al., 2021). It should also be noted that not all patients suffering from varicocele or pelvic congestion syndrome exhibit imaging features of left renal vein compression. Developing efficient collateral circulation can reduce pressure within the LRV (Ananthan et al., 2017; Gavrilov et al., 2024; Gulleroglu et al., 2022).

In patients with NCS, the angle of the superior mesenteric artery branching from the aorta is abnormally small, which may also underlie the superior mesenteric artery syndrome. This disorder involves the compression of the duodenum between the superior mesenteric artery and the aorta, causing symptoms such as nausea, vomiting, postprandial abdominal pain, or even features of acute abdomen. The coexistence of both syndromes is rare, but it can occur due to a similar formation mechanism (Brogna et al., 2023; Pacheco et al., 2023; Shi et al., 2019). The literature also describes clinical cases of patients where compression of the left renal vein was accompanied by other less frequently described symptoms, such as headaches associated with congestion of the epidural venous plexus resulting from the developed collateral circulation Devcic et al., (2022) arterial hypertension, Azhar et al., (2019), Wang et al., (2021b) orthostatic proteinuria in children Gulleroglu et al., (2022), Park et al., (2002) or fatigue (Cioffi et al., 2022).

Additionally, there have been sporadic cases where compression of the left renal vein led to severe complications such as thrombosis of the LRV Lizama et al., (2020), Nakashima et al., (2020), pulmonary embolism Hori et al., (2021) or left renal vein aneurysm (Lazar et al., 2019). The exact impact of the degree of LRV compression on left kidney function is not well understood. Studies conducted by Gavrilov et al., (2024) indicate that compression of the left renal vein without hematuria or proteinuria does not significantly affect the function of the left kidney. However, more research is needed due to the absence of patients with the symptoms mentioned above in the study (Gavrilov et al., 2024).

An important syndrome worth mentioning is the loin pain hematuria syndrome (LPHS), which presents with hematuria and chronic loin pain. Its exact etiology is unknown, but it is postulated that NCS may initiate the process leading to the development of LPHS. It should also be noted that the signs of this syndrome persist even after the compression on the LRV is relieved (Campsen et al., 2021). Due to the fact that LPHS and NCS may present with similar symptoms, it is suggested that a thorough differential diagnosis be performed when suspecting any of those two diseases (Zubair et al., 2016).

Epidemiology

NCS is qualified as a rare disease, although its prevalence is not entirely known. It is supposed to differ between different age groups. Very likely, male patients are the ones to be diagnosed earlier, and for both sexes, the peak of the disease's diagnosis lies within the second or third decade of life (Orczyk et al., 2016; Ribeiro et al., 2020; Penfold et al., 2024). Occurrence between males and females is, however, still not well established, given that some studies have shown that the compression of the left renal vein is more common in women Ribeiro et al., (2020), Góes et al., (2020) while others do not find any differences in occurrence between the thought (Yoon et al., 2022; Grimm et al., 2013).

New studies suggest that NCS and NCP might be much more prevalent than previously thought (Yoon et al., 2022; Grimm et al., 2013). What is more, discrepancy within different diagnostic criteria results in overdiagnosing of NCP during the imaging examination, while at the same time, NCS is undiagnosed due to nonspecific symptoms (Penfold et al., 2024). Posterior NCS is less frequent than anterior NCS, and it is postulated that the prevalence is the same for men and women. The variation of posterior NCS with singular retroaortic vessel occurs more frequently and more often causes symptoms in patients, compared with the variation with doubled renal vein, where one vessel passes in front of the aorta and the other behind it (Park et al., 2018; Shi et al., 2018).

Diagnosis

Due to nonspecific symptoms, diagnosing NCS can be challenging. Additionally, it is essential to remember that radiological features of LRV compression do not always cause clinical symptoms, necessitating thorough differential diagnosis to accurately pinpoint the cause of the symptoms. Another reason for diagnostic difficulty is the previously mentioned lack of consensus on radiological criteria (Ribeiro et al., 2020). Phlebography, along with intravascular pressure measurement, is considered the "gold standard". However, due to the invasive nature of the procedure, it is used as a last resort and is sometimes omitted in the diagnostic process (Ananthan et al., 2017; Wang et al., 2021b; De-Macedo et al., 2018). Currently, it is unclear whether the criteria used to diagnose Nutcracker Syndrome should differ between children and adults (Kurklinsky and Rooke, 2010).

Radiological features observable in US, CT, and MRI include:

Anatomical abnormalities, atypical vessel courses, and local changes that may cause compression of the left renal vein and features of increased collateral circulation (Englund and Rayment, 2018).

Decreased aortomesenteric angle, which is defined as the angle of branching of the superior mesenteric artery from the abdominal aorta (Figure 2). The angle values and cut-off points vary depending on the study. Some authors assume that the typical value of this angle ranges from 38 to 65 degrees, while an angle below 25 degrees is considered a cut-off point (Granata et al., 2021; Rangel-Villalobos et al., 2022). Other studies report a standard angle value ranging from 45 to 90 degrees Kim, (2019) and indicate that a value of 40 degrees or 41 degrees Said et al., (2013) is responsible for the onset of symptoms, with 35 degrees being a definitive diagnosis (Granata et al., 2021; Cioffi et al., 2022; Penfold et al., 2024; Velasquez et al., 2018). It should be noted that this angle changes with the patient's position, which presents another difficulty in diagnosis (Kolber et al., 2021; Gulleroglu et al., 2022). There are reports that in pediatric patients with orthostatic proteinuria, a decrease in the aortomesenteric angle ratio between upright and lying positions may be helpful in diagnosing NCS (Gulleroglu et al., 2022).

The ratio of the dilated part of the left renal vein to its narrow part: The vessel diameters are measured at the renal hilum and in the aortomesenteric part (Hangge et al., 2018; Cheon et al., 2006). A ratio greater than 4.9 has a strong predictive value (sensitivity 66.7%, specificity 100%) (Kurklinsky and Rooke, 2010; Hangge et al., 2018; Ananthan et al., 2017; Cioffi et al., 2022; Velasquez et al., 2018). Other values mentioned in the literature are >4. 5 Rangel-Villalobos et al., (2022),> 4.2 for children, and > 4.0 for adults (Penfold et al., 2024). Diagnostic difficulties and variations in establishing a uniform value for this ratio may partly result from the type of examination itself. In ultrasound (US), it is essential to remember the pressure of the US probe, which can affect the measured diameters (Kim, 2019). Additionally, the length of the LRV ranges from 6 to 10 cm, with a diameter of 4 mm to 5 mm (Kurklinsky and Rooke, 2010).

The distance between the superior mesenteric artery and the aorta: Less than 8 mm (Rangel-Villalobos et al., 2022).

Comparison of algorithms

In the course of the last few years, several algorithms have been presented for diagnosing and treating NCS. The common point between those algorithms seems to be a gradual approach to introducing invasive procedures (Nastasi et al., 2022; Ananthan et al., 2017; De-Los-Reyes et al., 2021; Meyer et al., 2022). All suggested diagnostic algorithms begin with the suspicion of NCS based on clinical symptoms and exclude other causes. To exclude these other causes, Nastasi et al. recommend conducting tests such as biochemistry, hematology, urinalysis, and, if necessary, cystoscopy (Nastasi et al., 2022). On the other hand, Chait et al. recommend performing cystoscopy in all patients with macroscopic hematuria (Chait et al., 2021).

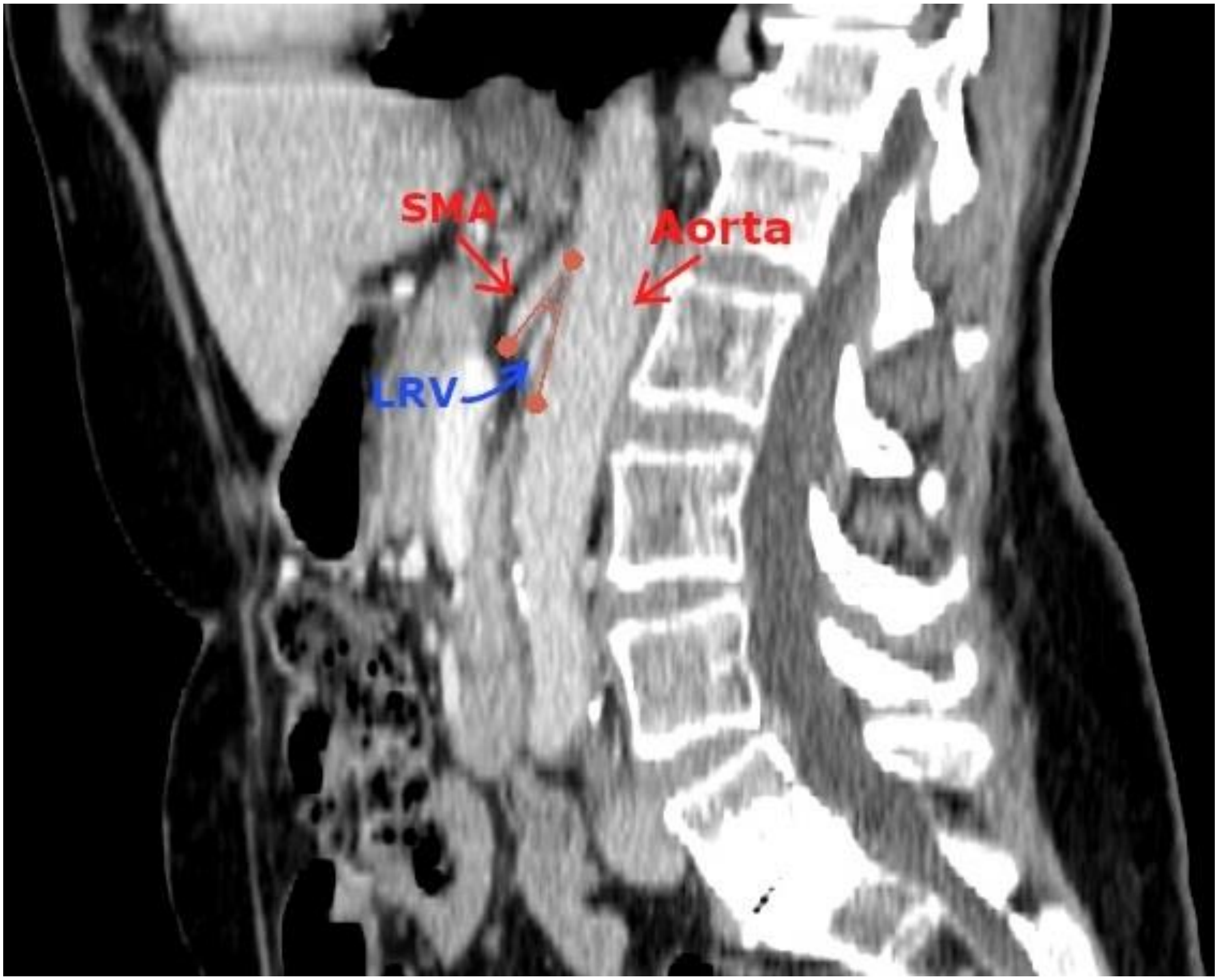

Figure 2 CT sagittal plane. The structures are labeled as follows: "LRV"-Left Renal Vein, "SMA" – Superior Mesenteric Artery. This image marks the angle between the aorta and the Superior Mesenteric Artery (indicated in pink; the angle is approximately 22.8°).

The first imaging test recommended by all algorithms is ultrasound (US). Nastasi et al., (2022) recommend performing angio-CT or angio-MRI along with US. In contrast, Ananthan et al., (2017) and De-Los-Reyes et al., (2021) recommend additional imaging tests when diagnostic difficulties are associated with the US examination. The algorithm presented by Meyer et al., (2022) designed for children, aims to avoid excessive testing and recommends proceeding directly to conservative therapy when both typical clinical symptoms and sonographic findings are present. In cases of further progression or persisting symptoms, Meyer et al., (2022) recommend angio-MRI and, as a surgical intervention, LRV transposition.

In cases of ongoing diagnostic difficulties, Nastasi et al., (2022), De-Los-Reyes et al., (2021) and Ananthan et al., (2017) recommend venography with intravascular pressure measurement. Additionally, De-Los-Reyes et al., (2021) suggest performing the University of Wisconsin-Loin Pain Hematuria Syndrome (UW-LPHS) test to confirm LPHS. In summary, there seems to be a consensus among the authors on using the US as the first imaging test and, in case of an uncertain diagnosis, CT and MRI. Venography may be helpful before surgical procedures (Nastasi et al., 2022). For children, the authors suggest minimizing the number of necessary tests.

Ultrasound (US)

The advantages of this method include a short examination time, non-invasiveness, and the absence of exposure to ionizing radiation. The main drawback is the requirement to be fasting before the test. US is the most commonly chosen type of examination in diagnosing

NCS (De-Macedo et al., 2018; Meyer et al., 2022). All the diagnostic algorithms compared above recommend performing the US as the first imaging test. Both B-mode and Doppler US are used in diagnostics. According to estimates, the sensitivity of Doppler US ranges from 69% to 90%, while specificity ranges from 89% to 100% (Kurklinsky and Rooke, 2010). In Doppler US, we can look for the ratio of peak blood flow velocity between the aortomesenteric segment and the renal hilum. The cut-off point for children is >4.7 (Cioffi et al., 2022). Other values include >5.0 for adults (sensitivity 80%, specificity 94%) and >4.7 for children (sensitivity 100%, specificity 90%) Kim, (2019) or a range from >4.2 to 5. 0 (Velasquez et al., 2018).

CT and MRI

CT or MRI is recommended as second-line imaging tests in all the compared algorithms. However, they have their drawbacks. MRI requires a more extended examination time but does not expose the patient to ionizing radiation and contrast. For this reason, MRI may be used as an alternative to CT in children (Atasoy et al., 2021; Wang et al., 2021a). In CT or MRI, we can observe "the beak sign", which is typical for NCS (Figure 3). It is defined as the angle formed by extending the anterior and posterior walls of the renal vein at the site of compression in the axial plane (Kim et al., 2011; Chait et al., 2021). The suggested cut-off value is 32 degrees. Studies show that the sensitivity of this sign is 91.7%, and the specificity is 88.9% (Kim et al., 2011; Hangge et al., 2018; Said et al., 2013; Kim et al., 2021).

In angio-CT, Kim et al., (2021) described the "jetting sign". This sign involves the rapid inflow of contrasted blood from the LRV into the inferior vena cava. It is characterized by the appearance of contrast-enhanced blood in the anterior part of the vessel or its spiculate outline in the late arterial phase. Additionally, Kim et al., (2021) described the "dilatation ratio", which is the ratio of the diameter of the LRV at the compressed site in the delayed and arterial phases. The jetting sign showed a sensitivity of 84.6% and a specificity of 71.9%, while the dilatation ratio had a sensitivity of 84.6% and a specificity of 63.2% for a cut-off point of 1.7 (Kim et al., 2021). In MRI, the use of T2- TRUFI, FSE T2WI, T2-HASTE, out of-phase T1, and T1-VIBE are highly useful in assessing the LRV (Kolber et al., 2021; Atasoy et al., 2021; R. Wang et al., 2021a).

Venography combined with intravascular blood pressure

The "gold standard" of diagnosis in NCS is claimed to be venography combined with intravascular blood pressure measurement. However, due to its high invasiveness, it is not chosen very often on the diagnostic path (Atasoy et al., 2021; Wang et al., 2021b). All algorithms compared above suggest performing this test in case of nondefining Doppler US, CT, or MRI results. Additionally, Meyer et al., (2022) do not include performing it on children in their diagnostic algorithm. Venography combined with intravascular blood pressure measurement relies on introducing an intravascular catheter and measuring blood pressure in the inferior vena cava and the left renal vein.

If the pressure difference exceeds 3 mmHg, it suggests an uncompensated form of NCS. It should be noted that the pressure difference may be less than 3 mmHg in individuals with well-developed collateral circulation (Kim et al., 2011; Cioffi et al., 2022; De-Los-Reyes et al., 2021). There are also suggestions to set the cut-off point at values greater than 2 mmHg or even greater than 1 mmHg (Nastasi et al., 2022; Velasquez et al., 2018). Moreover, the pressure difference may also be greater than 3 mmHg in some healthy individuals (Ananthan et al., 2017).

Other diagnostic tools

Other diagnostic measures rarely described in the literature include renal biopsy, cystoscopy, intravenous pyelogram (IVP), arteriography, scintigraphy, and intravascular ultrasound (IVUS) (Park et al., 2018; Meyer et al., 2022). Additional diagnostic exams can be very useful in unresolved cases causing diagnostic issues and as a tool for expanded differential diagnosis.

Treatment

Two approaches can be taken to treat the Nutcracker Syndrome: Interventional or conservative. Many authors recommend the conservative one as the first-line strategy in patients with mild symptoms. For patients presenting with severe symptoms such as significant hematuria, intense flank pain, or when symptoms persist, interventional treatment is recommended (Park et al., 2018; De-Macedo et al., 2018; Stawiarski et al., 2017). Choosing the optimal interventional treatment method is not straightforward. It should be individualized, related to the cause of LRV compression, and based on an analysis of the advantages and disadvantages of each therapeutic method.

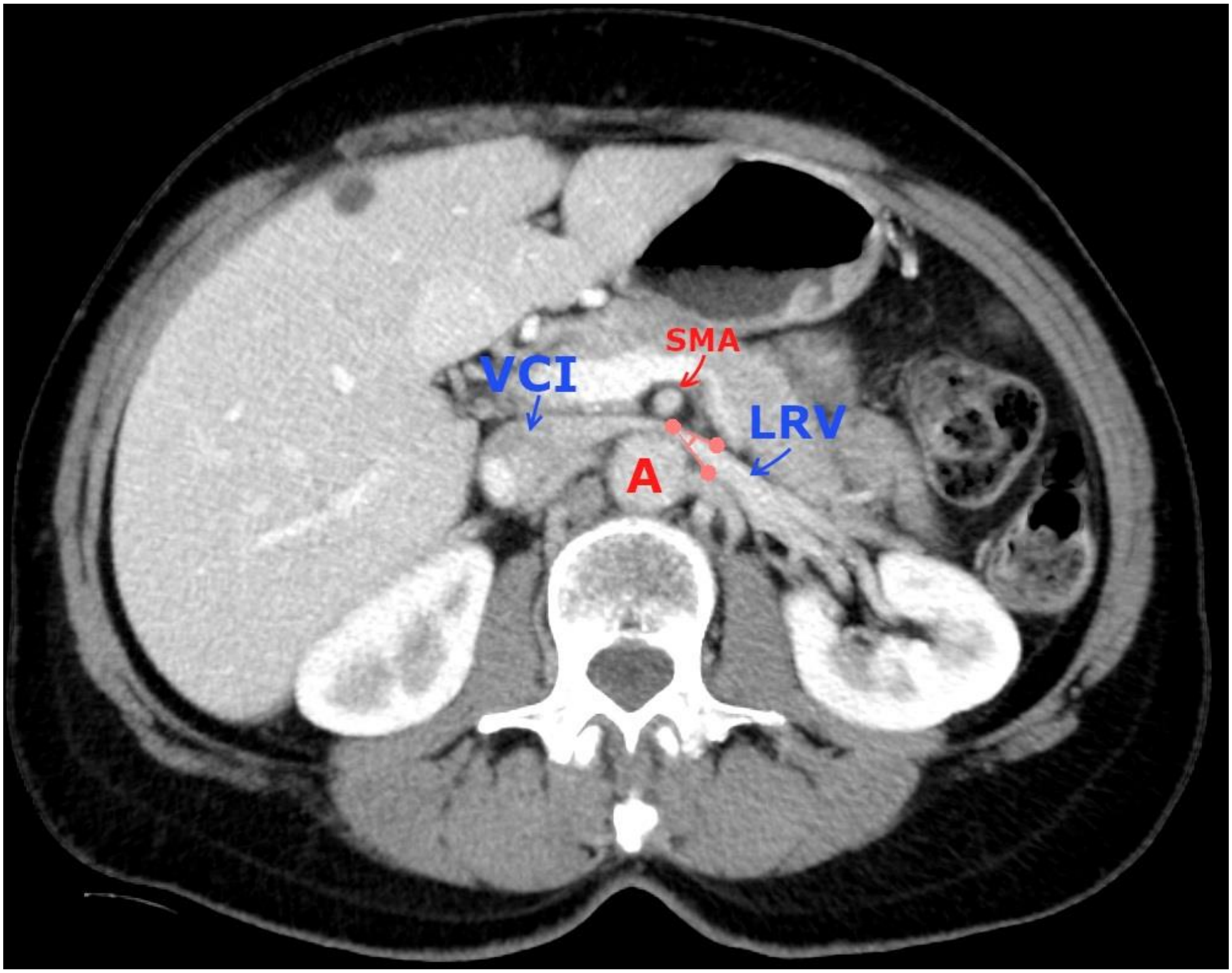

Figure 3 CT axial plane. The structures are labeled as follows: "A"-Aorta, "LRV"-Left Renal Vein, "SMA" – Superior Mesenteric Artery, "IVC"-Inferior Vena Cava. The "beak sign" is also visible (indicated in pink; the angle is approximately 29.5°).

Currently, laparoscopic surgery is gaining attention and being used more often (Kolber et al., 2021; Fuentes-Perez et al., 2023). The most commonly performed surgeries include left renal vein transposition (LRVT) (Ananthan et al., 2017; De-Macedo et al., 2018). For patients with posterior NCS, anterior open LRVT is recommended (De-Macedo et al., 2018). Other methods, such as intravascular or combined intravascular and extravascular procedures, have also been described in patients with posterior NCS (Park et al., 2018; Stawiarski et al., 2017). Robot-assisted laparoscopic procedures have also been reported (Thaveau et al., 2015).

Conservative treatment

A wait-and-see strategy seems particularly useful in children, for whom the recommended observation period is 24 months. In a group of patients under 18 years of age described by Meyer et al., (2022) 42.8% experienced complete symptom resolution with conservative treatment, and 52.2% showed partial clinical improvement. The percentage of children whose symptoms persisted was 5% (Meyer et al., 2022). The exact cause of symptom resolution is unknown. It is postulated that spontaneous remission may be related to growth, weight gain, collateral circulation development, increased fat and connective tissue (Kolber et al., 2021; Ananthan et al., 2017; Cioffi et al., 2022; Meyer et al., 2022). To minimize proteinuria, ACE inhibitors or ASA may be considered (Kolber et al., 2021; Ananthan et al., 2017; Meyer et al., 2022). The recommended adult observation period is 6 months (He et al., 2014; De-Macedo et al., 2018).

Endovascular Treatment

The methods used in endovascular treatment include stenting, balloon angioplasty, or embolization. Endovascular methods are gaining popularity (De-Macedo et al., 2018). Their use has been described for both adults and children. Among some authors, endovascular stenting in minors raises controversies related to introducing a stent into a not fully-developed body (De-Macedo et al., 2018; De-Los-Reyes et al., 2021). Endovascular stenting is one of the less invasive methods. It involves placing a stent inside the LRV to reduce vessel compression. The advantages of endovascular treatment include reduced procedure invasiveness, local anesthesia use, and a shorter hospitalization time (De-Macedo et al., 2018; Fuentes-Perez et al., 2023). During the procedure, IVUS can be very helpful, as it allows for assessing the degree of compression and the site of the greatest pressure and helps select the appropriate stent size (Cronan et al., 2021).

In the group of patients analyzed by Fuentes-Perez et al., (2023) who underwent endovascular stenting, 76% experienced symptom relief. It should also be remembered that the use of this method is associated with the need to take antiplatelet medications such as clopidogrel or ASA for around 2 to 3 months in the adult population and up to 6 months in the pediatric population (Stawiarski et al., 2017; Cronan et al., 2021). Severe complications of endovascular stenting include vessel damage, in-stent thrombosis, erosion, and stent migration (Stawiarski et al., 2017; Cronan et al., 2021; Wu et al., 2016). To reduce the risk of stent migration, the implant is typically oversized by approximately 20% (Stawiarski et al., 2017). Additionally, cases have been described of simultaneous stenting and embolization of dilated gonadal vessels to treat varicocele or PCS (Chait et al., 2021; Aghdasi et al., 2022).

Transposition of the Left Renal Vein

Transposition of the left renal vein is one of the most common and effective surgical procedures and is considered the “gold standard” in the surgical treatment of NCS. It is a procedure that involves detaching the LRV from the inferior vena cava and then suturing it distally to its original location (De-Macedo et al., 2018; Said et al., 2013; Hartung et al., 2010). It can be performed during open surgery, laparoscopy, or robotic surgery (Reed et al., 2009; Thaveau et al., 2015; Hartung et al., 2010). Prolonged compression on the vessel can cause its deformation, which sometimes necessitates the use of a patch, cuff, or vascular prosthesis during surgery (Said et al., 2013; Zhang et al., 2015). This procedure can carry complications, including restenosis, thrombosis, or bleeding (Penfold et al., 2024). It is a method recommended by Meyer et al., (2022) for the treatment of NCS in children.

Extravascular Stent Placement

Extravascular stent placement is a relatively new but effective method that involves the implantation of an extravascular stent, which provides support to relieve the compression of the left renal vein (Fuentes-Perez et al., 2023). The use of 3D printing has been described as a way to create individualized stents (Fuentes-Perez et al., 2023; He et al., 2022). This procedure is more invasive and requires general anesthesia, but unlike the endovascular approach, it eliminates the need for anticoagulant medications (Fuentes-Perez et al., 2023). In some cases, prolonged vein compression caused the formation of a fibrous ring, the cutting of which was proven crucial for achieving a good therapeutic effect with this method (Chen et al., 2019).

Other treatment options

Other alternative methods described in the treatment of NCS include:

The hybrid approach is a relatively new and advanced method for treating NCS It involves the surgical transposition of the LRV followed by the insertion of a stent into the vessel to further reduce compression (Chait et al., 2021; Stawiarski et al., 2017).

Kidney auto-transplantation is a rarely performed and challenging method. It involves nephrectomy followed by transplanting the kidney into the iliac fossa (Ananthan et al., 2017; Penfold et al., 2024). This method also seems to help patients with LPHS (Campsen et al., 2021). The University of Wisconsin-Loin Pain Hematuria Syndrome (UW-LPHS) test can help to identify patients for whom this procedure might be effective (Sollinger et al., 2018).

Gonadal vein transposition is an effective, alternative surgical method for treating NCS (Gilmore et al., 2021). It uses the gonadal vein, which is then anastomosed, to the external iliac vein, the inferior vena cava, common iliac vein or the inferior epigastric vein (Gilmore et al., 2021; Debucquois et al., 2021; Li et al., 2020). Such a connection allows blood to flow from the LRV through the gonadal vein. An existing dilated gonadal vein is a prerequisite for this surgery (Debucquois et al., 2021).

Superior mesenteric artery transposition is an effective but rarely described procedure that involves moving the superior mesenteric artery to reduce the pressure it exerts on the LRV. This procedure is more complex than LRV transposition and can be associated with mesenteric ischemia (Kolber et al., 2021; Yang et al., 2012).

Saphenous vein bypass – In this method, the great saphenous vein is used to create an additional connection, thus facilitating blood flow between the LRV and the IVC (Said et al., 2013).

In extreme cases, nephrectomy has also been described to reduce persistent symptoms that do not respond to other treatment methods (Ananthan et al., 2017).

Despite the NCS being known for a long time, it has still not been entirely discovered. The exact symptoms-provoking pathomorphological abnormalities caused by congestion are not precisely defined. Moreover, a connection between LPHS and NCS is also not precisely known. There is a lack of consensus on diagnostic criteria, especially on cut-off values. Another aspect of NCS that is not entirely understood is whether the diagnostic criteria should be the same or different for the pediatric population. As a result of all the mentioned inconsistencies, there isn't clarity about the actual prevalence of NCS, and there is a high probability of NCP being overdiagnosed and NSC being underdiagnosed.

On the other hand, because of its rare occurrence, some patients may experience difficulties on the diagnostic path. Symptoms of various diseases can also overlap, making it necessary to perform a thorough differential diagnosis. Another inconsistency in diagnostic criteria refers to the selection of diagnostic exams. The choice of an appropriate treatment may also not be obvious. Although the transposition of the left renal vein is still known to be the "golden standard" of NCS treatment, other methods, such as extravascular or intravascular treatment, are possible alternative ways of treatment. More research is still needed to compare and precisely evaluate the effectiveness and complications of various NCS treatment methods.

4. CONCLUSION

Nutcracker Syndrome manifests through hematuria, lumbar pain, or proteinuria. Other less common symptoms include varicocele, hypertension, or pelvic congestion syndrome (PCS). NCS arises from the compression of the left renal vein, and it can be divided into anterior NCS, where the vessel is compressed between the aorta and the superior mesenteric artery, and the rarer posterior NCS, where the LRV is compressed between the spine and the aorta. The situation in which clinical symptoms do not accompany the existing vessel compression is referred to as the Nutcracker Phenomenon. The exact prevalence of the syndrome is unknown, but due to its nonspecific symptoms, it may be underestimated. Various diagnostic algorithms have recently been proposed to facilitate the diagnosis process.

However, the lack of uniform cut-off values in imaging studies complicates the definitive diagnosis. There are many effective methods for treating NCS, including both conservative and surgical approaches. The most commonly used treatments include LRV transposition and endovascular stenting. New alternative and effective treatment methods, such as extravascular stenting and hybrid surgeries, have been described in recent years. Despite these advancements, NCS is still not fully understood. The exact impact of blood congestion on the kidney, the mechanism causing symptoms, and its long-term effect on kidney function are still unknown. There is also no consensus on whether diagnostic values for children can be applied to adults. More research is needed to gain a better understanding of the syndrome.

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Author Contributions

Conceptualization: PW and KW; Data curation: AG, AS, JS, DB; Writing - rough preparation: PW; Writing - review and editing: GL and KW; Visualization: ND, MN; Supervision: GL and MW; Project administration: PW. All authors have read and agreed with the published version of the manuscript.

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The authors declare that there is no conflict of interests.

Data and materials availability
All data sets collected during this study are available upon reasonable request from the corresponding author.

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