

Role of physical exercise in an overlooked nutcracker syndrome occurred in a patient with diaphragmatic relaxation: a case report

Nefrologo in corsia

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ABSTRACT

Background: Nutcracker syndrome (NCS) is caused by extrinsic compression of the left renal vein (LRV), usually between the abdominal aorta (AA) and superior mesenteric artery (SMA). This rare disease includes symptoms such as hematuria, left flank pain or abdominal pain, varicocele in males, proteinuria, anemia, gynecological symptoms (dyspareunia, dysmenorrhea).

Case report: We report the case of a 48-year-old female patient, who experienced left abdominal colic after intensive physical exercise, finally resulting in a diagnosis of NCS. This abdominal pain was disabling for daily activities, it was controlled by analgesic drugs and led to hospital admissions. In-depth examinations were recommended to the patient to investigate the etiology of these attacks. A bad rotated and ectopic left kidney, which was located superior to the spleen, at the level of the left hemithorax base, was found due to the presence of a diaphragmatic relaxation in the posterior area, which caused an upward displacement of the kidney, part of the colon and omental fat. Because of the presence of a compression of the LRV by the SMA and the AA, the nephrologist diagnosed a NCS, presenting with abdominal pain following physical exercise, proteinuria and dysmenorrhea. Conservative treatment was chosen for the patient.

Conclusions: The patient was recommended to engage in a moderate and regular physical activity, avoiding acute and intense exercise: hypopressive abdominal gymnastics was suggested. The role of physical exercise in triggering painful attacks and its role in rehabilitation to prevent the same attacks was crucial for the patient.

KEYWORDS: physical exercise, nutcracker syndrome, abdominal pain, diaphragmatic relaxation

Introduction

Left renal vein entrapment syndrome, or nutcracker syndrome (NCS), is caused by extrinsic compression of the left renal vein (LRV), usually between the abdominal aorta (AA) and superior mesenteric artery (SMA) (anterior NCS), or, rarely, between AA and spinal column (posterior NCS) [1]. It is a rare disease with an unknown prevalence, occurring more frequently in females in their thirties and forties [2, 3]. Renal veins are large vessels anterior to the renal arteries, which flow into the inferior vena cava almost at right angles. The LRV is three times longer than the contralateral and it is posterior to the lienal vein and body of the pancreas. The vessel crosses the anterior surface of the AA, just inferior to the origin of the SMA. The left genital vein enters the LRV from below, while the left adrenal vein flows into it approximately at the level of the midline. A study enrolling 112 patients reported common NCS typical symptoms including hematuria (78.57%), left flank pain (38.39%), varicocele in males (35.71%), proteinuria (30.36%), and anemia (13.39%) [4]. Abdominal pain, gynecological symptoms (dyspareunia, dysmenorrhea), orthostatic hypotension, fatigue and infertility are also been described [5, 6]. Autonomic dysfunction symptoms such as hypotension, syncope, nausea and weight loss may occur [7]. Diagnosis must be reached by exclusion and by a stepwise approach including anamnesis, physical examination, instrumental methods such as doppler ultrasonography (DUS), computed tomography angiography (CTA), magnetic resonance imaging (MRI), venography, and intravascular ultrasound (IVUS) [3]. Table I shows clinical conditions to be considered in differential diagnosis with NCS.

Clinical conditions for differential diagnosis with NCS [3]	
Pancreatic/retroperitoneal/renal tumor	Nephrolithiasis
Para-aortic lymphadenopathy	Pyelonephritis
Abdominal aortic aneurysm	Left renal ptosis
Overarching testicular artery	Lordosis
Fibrolymphatic tissue between the aorta and SMA	Reduced retroperitoneal/mesenteric fat
High disposition of the LRV	Gravid uterus compressing the renal vasculature

Table I: clinical conditions to be considered in differential diagnosis with nutcracker syndrome [3]. NCS: Nutcracker Syndrome; SMA: superior mesenteric artery; LRV: left renal vein

About imaging, a reno-caval pullback gradient ≥ 3 mmHg, a maximum flow velocity five times higher than normal values in the LRV passing the SMA compared to the renal hilum, or an angle less than 45 degrees between the AA and SMA on CTA or MRI are considered as diagnostic criteria [8]. Conservative treatment, endovascular treatment or open surgery approach depend on NCS symptoms and their severity.

Physical activity is recommended by Health Authorities leading to a real health benefit for the body and regular exercise can mediate a protective effect, thanks to the stimulation of anti-inflammatory molecules and the reduction of visceral fat. Increased oxidative stress, weight and fat gain, low-grade systemic inflammation and risk of developing diseases such as type 2 diabetes, cardiovascular disease and some cancers are associated with physical inactivity [9]. The role of physical activity is undoubtedly positive in physiological and pathological conditions, but certainly the type of exercise and the workload should be personalized in light of the characteristics of the subject and his anamnesis. We report the case of a female patient, who experienced left abdominal colic after intensive physical exercise, finally resulting in a diagnosis of an anterior NCS (Figure 1).

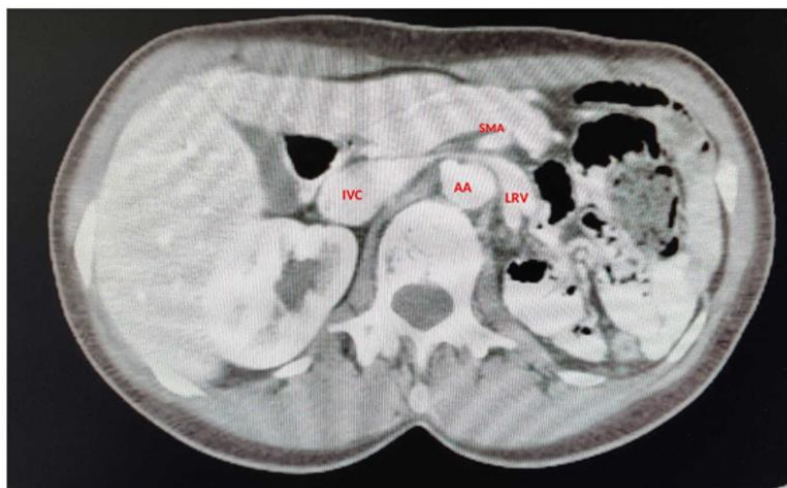


Figure 1: CT shows the compression of the left renal vein between the aorta and superior mesenteric artery in a patient affected by nutcracker syndrome. LRV: left renal vein, AA: abdominal aorta, SMA: superior mesenteric artery, IVC: inferior vena cava.

Case report

A 48-year-old woman, apparently in good health, suffered from left abdominal colic after practicing several hours of strenuous physical exercise. The woman practiced bodyweight aerobic exercises, gym exercises, and abdominal flexion and extension exercises several times a week. This abdominal pain was disabling for daily activities and was controlled by statim administration of analgesic drugs (e.g. intramuscular ketorolac) and also led to hospital admissions. The patient was within the normal weight range (BMI 20.74, normal range: 18.5-24.99) and suffered from pharmacologically controlled mild hypertension for about five years. In addition, the patient was suffering from anxious depressive syndrome and dysmenorrhea. Her therapy involved the use of nebivolol, olmesartan cilexetil and estradiol/progesterone. Routine blood chemistry tests had been performed multiple times and were normal. However, the latest analyses showed marked proteinuria (2 g/day) and an *Escherichia Coli* infection. Then antibiotic therapy was started, which led to the eradication of the infection; unfortunately, the episodes of abdominal colic increased their frequency. At this point, the patient underwent a urological examination, which suggested the possibility that a urinary calculosis could trigger the pain attacks, although not identifiable by ultrasound exams. The renal ultrasound, on the other hand, showed the ectopy of the left kidney displaced inferomedially about 1-2 cm superior to the umbilical scar. A therapy based on hydrochlorothiazide and food supplements based on Potassium Citrate, Magnesium, and Zinc was prescribed to stem the clinical picture, and a specific diet low in calcium was started. After two months since these dietary and therapeutic regimens have started, the frequency and intensity of abdominal colic remained unchanged. At this point, in-depth examinations were recommended to the patient to investigate the etiology of these attacks. First, the patient performed a chest and abdomen CT scan, which showed an asymmetry of the pulmonary fields due to a suspected left posterolateral hernia (Bochdalek's hernia) with evidence of intestinal loops (Figure 2); in addition, there was a dislocation of both the spleen and the left kidney, which appeared to be badly rotated. The CT urography also showed a badly rotated and ectopic left kidney, which was located superior to the spleen, at the level of the left hemithorax base, due to the presence of a diaphragmatic relaxation in the posterior area which caused an upward displacement of the kidney, part of the colon and omental fat. Then the patient underwent a CT angiography which showed the presence of a compression of the LRV, due to a compression exerted by the SMA and the AA, reporting an angle of 30 degrees (normal value >45 degrees). The

MRI also confirmed this anatomical picture. The nephrologist diagnosed a NCS, presenting with abdominal pain following physical exercise, proteinuria and dysmenorrhea. Conservative treatment was chosen for the patient, considering the syndromic picture and age. The patient was recommended to engage in a moderate and regular physical activity, avoiding acute and intense exercises; a dietary regimen was established aimed at the formation of an adipose panniculus that would restore normal anatomical relationships. The patient is continuing to be followed up periodically by the nephrologist, to ascertain the evolution of the case both from a clinical and laboratory point of view (at a six-month follow-up, the proteinuria was 0.4 g/day).

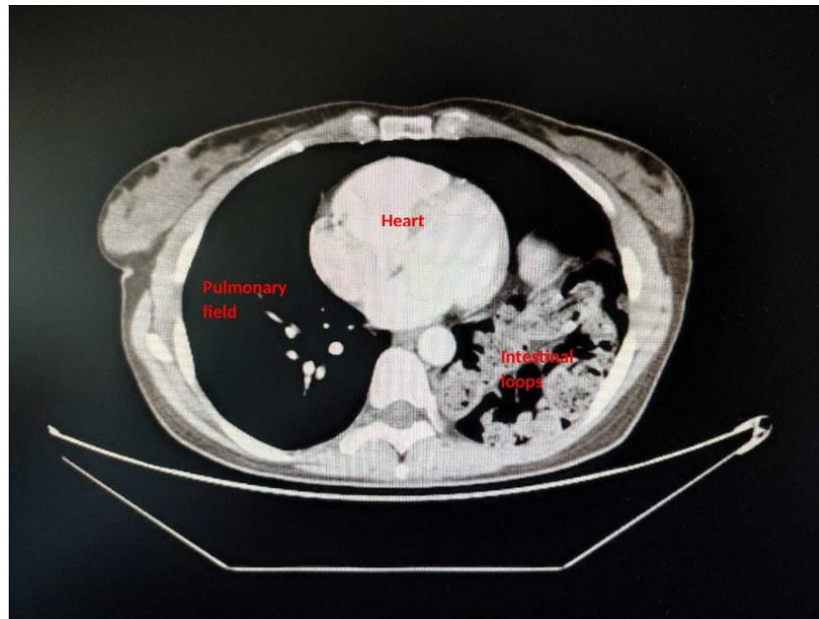


Figure 2: CT scan showed an asymmetry of the pulmonary fields due to a suspected left posterolateral hernia (Bochdalek's hernia) with evidence of intestinal loops.

Discussion

This article reports the case of a patient who didn't show any specific multiple symptoms so that the diagnosis was delayed despite the patient had been followed several times from a clinical point of view. NCS was eventually diagnosed, a rare disease whose diagnosis is often unknown due to the low frequency of cases. Furthermore, the syndrome occurred in an equally particular and atypical case, given that the patient's left kidney was ectopic and the topographical anatomy of the left abdomen was disturbed due to the presence of an infrequent anatomical defect of the diaphragm and suspected herniation of the viscera.

The diagnostic process was long and difficult also due to the absence of typical symptoms, such as hematuria [1]. The patient also was thin with a weight of only 62 kg and belonged to low limit of the normal BMI range. Among conservative treatments weight gain was recommended, because an increase in retroperitoneal adipose tissue has demonstrated to resolve NCS symptoms in 30% of cases by a reduction in LRV tension [10]. The abdominal colic was triggered by intense physical exercise, especially as a result of exercises that strained the abdominal muscles and increased intra-abdominal pressure. It is thanks to the physical exercise practiced by the patient that the painful symptoms were triggered, which prompted the patient to investigate the cause and the diagnostic process to be concluded. Some exercises increase intra-abdominal pressure such as abdominal exercises (e.g. sit-ups, curl-ups, crunches, double leg lifts, exercises on machines), weighted rotation exercises (e.g. seated medicine ball twists, suspended torso rotations, rotating planks) and planks

on toes and its variations (e.g. planks with feet on ball or in suspension straps, planks on hands and toes and moving to elbows). In the scientific literature, it is known that intra-abdominal pressure associated with exercise can have a negative effect on pelvic floor muscle, even if the damage varies among activities and among women [11]. Repetitive, high-impact sports seem to expose females to the highest risk for urinary incontinence [12]. In a comparative *in vivo* study treadmill exercise determined an increase in intra-abdominal pressure causing inhibition of venous return by compression of the large abdominal veins [13]. On the other hand, it has always been known that physical exercise benefits patients with chronic renal disease by improving blood pressure and their maximal oxygen consumption level [14]. As also presented in our case, hypertension also appears among the symptoms of NCS. This symptom was reported in the case of NCS in a young Japanese woman by Hosotani et al. and in a 22-year-old Chinese man who showed left flank pain after activity [15, 16]. Our patient was used to performing intense physical exercise; therefore, it would be desirable not to upset her habits. To make sure that physical activity couldn't trigger any attacks of abdominal pain, she should perform exercises at a moderate intensity, avoiding exertion. The most suitable training method for the patient could be hypopressive abdominal gymnastics (HAG), a global and systemic postural technique, which is becoming very well-known [17]. This training technique relaxes the diaphragm, reduces intra-abdominal pressure, and is already used for the treatment of abdominal diastasis in the postpartum period, urinary incontinence, pelvic prolapse and chronic low back pain. Caufriez, in the 80s, developed a training consisting of 33 hypopressive exercises to be performed in different postures (sitting, kneeling, quadrupedal and supine). Each exercise involved a hypopressive maneuver including a prolonged exhalation followed by apnea. During these exercises an intense and voluntary contraction of the inspiratory accessory muscles, such as the serratus anterior, the external intercostal, the scalene and the sternocleidomastoid, was performed, keeping the glottis closed (diaphragmatic suction). HAG produces direct activation of the transverse abdominal muscle, leading to a strengthening of the abdominal wall and stabilization of the spine [18]. The lumbar spine and hamstrings become more flexible and body posture reorganizes [19]. Clearly, in our case a conservative approach was chosen, but the patient needs a close follow-up that allows to monitor her conditions from a clinical and hemodynamic point of view.

Conclusions

The described case focuses on the difficulty of diagnosing a rare pathology such as NCS, which can only be recognized by differential diagnosis after excluding the most common causes of abdominal pain. Collateral symptoms must not be overlooked, and the patient's physiological and pathological history must be taken into consideration. Physical exercise can always be recommended, even in pathological conditions such as in this case; obviously type, duration and intensity of physical exercise should be adapted to the general status of the patient. Furthermore, a balanced diet could also benefit the patient. In our case and in those under conservative treatment it is useful and necessary to carefully monitor the patients to follow the evolution of the pathology.

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