

# A Rare Case of Bilateral Flank Pain: Anterior Nutcracker Syndrome and Post-pregnancy Hydronephrosis due to Adynamic Ureteral Segment

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## ABSTRACT

Since hydroureteronephrosis related to the adynamic ureter segment is mostly seen in childhood, it may be difficult to diagnose in adulthood. The entrapment of the left kidney vein most usually occurs between the abdominal aorta and the superior mesenteric artery (anterior nutcracker syndrome). A case of bilateral flank pain, anterior nutcracker syndrome and hydronephrosis, due to an adynamic ureter segment developing 1 year after giving birth is presented in this paper. According to our research, there is no case report of anterior nutcracker syndrome and hydronephrosis due to an adynamic ureter segment coexistence.

**Keywords:** Adynamic ureteral segment, anterior nutcracker syndrome, flank pain, hydronephrosis

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## INTRODUCTION

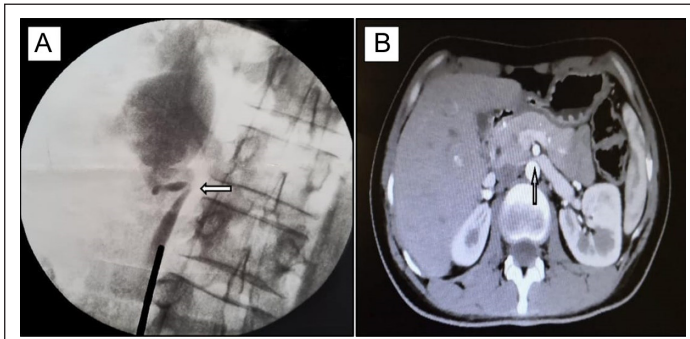
Urethrectasis can be congenital (mega-ureter) or acquired (hydro-ureter). Megaureters can be of reflux and nonreflux (obstructive) type. Primary nonreflux megaureters are caused by the presence of the prevesical adynamic segment causing a functional obstruction. The musculature of this segment is abnormal in both function and structure. It appears to be most commonly due to an abnormality or delay in the development of the muscle in the distal ureter adjacent to the ureterovesical junction at 20 weeks gestation.<sup>1,2</sup> The entrapment of the left kidney vein (LKV) most usually occurs between the abdominal aorta and the superior mesenteric artery (ANCS: Anterior nutcracker syndrome). These patients are usually managed conservatively with a focus on weight gain to increase retroperitoneal fat, which leads to decompression of the LKV due to a change in the position of the left kidney. If symptoms of significant left flank pain persist, reported interventions include endovascular stenting or open surgery with

LKV transposition but repeated interventions may be necessary.<sup>3,4</sup> There is not enough data on the relationship between these 2 pathologies in the literature, and according to our research, a case report in which both pathologies were presented together was not observed. For this reason, a case of bilateral flank pain; ANCS and hydronephrosis due to an adynamic ureter segment developing one year after giving birth is presented here.

## CASE PRESENTATION

A 32-year-old female patient presented with complaints of blunt and suffocating right flank pain that started 1 year after giving birth and lasted for an average of 5 to 7 days for 1 year and left flank pain, which had been present for many years, especially in the menstrual period. The patient's vitals, kidney function tests, and complete urinalysis were normal (creatinine: 0.69 mg/dL, blood urea nitrogen: 13 mg/dL; complete urinalysis: protein negative, leukocyte negative, erythrocyte negative). In addition, there is no history of any disease, operation other





**Figure 1.** Retrograde pyelogram (RPG) and computed tomography (CT) angiography samples of the patient. A: The curvature and stenosis of the right ureter on RGP is shown with arrow. B: Compression of the left kidney vein between the abdominal aorta and the superior mesenteric artery on CT angiography is marked with an arrow.

than cesarean section, and drug use. Urinary system ultrasound was evaluated in the direction of right ureteropelvic (UP) stenosis. Computed tomography (CT) of the entire abdomen was compatible with right UP stenosis. No stone, mass, or similar occlusive lesion was observed on CT. Subsequent magnetic resonance (MR) urography was interpreted as right UP stenosis. <sup>99m</sup>Tc-dimercaptosuccinic acid scintigraphy was evaluated as non-occlusive findings. A retrograde pyelogram (RGP) was then performed on the patient: A double-J stent (DJS) was also placed during the procedure to the patient, whose right ureter enlargement, curving, and narrow segment were observed in RGP (Figure 1A). Double-J stent was removed 2 months later, but the patient's complaints relapsed after the stent was removed. Thereupon, CT angiography was performed on the patient: ANCS was observed on the left (Figure 1B). No vascular pathology was observed on the right side. Because the patient could no longer bear the agony in her right flank, a right laparoscopic pyeloplasty was performed, and the adynamic section in the right ureter was removed. No intervention was made for ANCS since the patient's clinic was not severe and had left flank pain that became evident only during menstrual periods. In the 2-years follow-up of the patient, the complaint of right flank pain did not recur after the operation.

## MAIN POINTS

- In daily practice, there are many outpatient clinic presentations with complaints of flank pain. In this case, the togetherness of ureteropelvic stenosis and anterior nutcracker syndrome (ANCS), which are 2 rare causes of flank pain, is presented.
- It should be kept in mind that there may be an adynamic ureteral segment in UP stenosis detected without a space-occupying lesion (stone, tumor, etc.), especially in female adults who have just given birth.
- Anterior nutcracker syndrome should be considered in the differential diagnosis of a patient presenting with flank pain, especially in patients with low body weight and symptoms of pelvic congestion.

## DISCUSSION

In the literature, the words nutcracker phenomenon and nutcracker syndrome (NCS) are occasionally used interchangeably. The nutcracker phenomenon describes anatomical characteristics that are indicative of a nutcracker but do not cause clinical symptoms. Patients having clinical symptoms related to nutcracker anatomy are referred to as NCS.<sup>3</sup> There are 2 forms of LRV entrapment: anterior and posterior. The compression of the LKV between the abdominal aorta and the lumbar vertebral body is known as the posterior nutcracker phenomenon and is a rare condition. Anterior nutcracker syndrome, which is more frequently encountered in the literature, was seen in our case. The intensity of symptoms and their expected reversibility in relation to the patient's age and stage of the illness should guide treatment decisions in ANCS. Symptoms that are mild and acceptable can be treated conservatively. Recurrent gross hematuria with anemia, acute flank pain, kidney functional impairment, and inefficacy, or exacerbation of conservative treatment of chronic orthostatic proteinuria after 24 months of follow-up may necessitate surgery.<sup>5</sup> No intervention was made for ANCS in the present case since the patient's clinic was not severe; laboratory values were normal, and left flank pain became evident only during menstrual periods.

Hydroureteronephrosis associated with the adynamic ureteral segment is mostly observed in children, and data on its occurrence in adults are very limited. Barbalias et al<sup>6</sup> reported that four cases with a mean age of 45 were treated with a self-expandable intraureteral metallic stent.<sup>6</sup> Again, Ramalingam et al<sup>7</sup> reported 3 cases, the youngest, who was 16 years old, was successfully treated with laparoscopic Boari flap ureteric reimplantation. However, according to our research, there is no case report describing severe flank pain due to hydroureteronephrosis related to the adynamic ureter segment 1 year after giving birth, without any previous complaints, operation history, and comorbidities. In today's world, non-operative management is the favored method. In the following circumstances is surgical intervention recommended: considerable impairment of urine flow; worsening kidney function during the monitoring period; and recurrent urinary tract infection despite appropriate antibiotic prophylaxis.<sup>2</sup> In our case, surgical procedure was performed due to pain that impairs quality of life and stenosis preventing urinary flow.

## CONCLUSIONS

In conclusion, the ANCS should be kept in mind in female patients presenting with complaints of left flank pain, especially during menstrual periods. Adynamic ureteral segment should be considered in adult female patients presenting with the complaint of flank pain after pregnancy and non-obstructive hydronephrosis. The presented case was successfully treated with laparoscopic pyeloplasty and removal of the adynamic ureter segment.

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