Chronic pelvic pain: the role of the nutcracker syndrome

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ABSTRACT

Chronic pelvic pain is a problem at the same time common and underdiagnosed in women. Some literature reports show an incidence of up to 15% in women aged between 18-50 years, with impact over quality of life and economy. Among the causes of chronic pelvic pain, pelvic congestion syndrome stands out, characterized by pain, dysuria, hematuria, dysmenorrhea, dyspareunia and vulvar congestion, often accompanied by vulvar varices, described in 1949 by Taylor. We herein report a case of a patient with chronic pelvic pain in whom we diagnosed a nutcracker syndrome, characterized by stenosis of the left renal vein between the superior mesenteric artery and the aorta, with consequent status of left gonadal plexus hypertension, pelvic varices and symptoms of pelvic congestion. The treatment was varicose pelvic veins embolization, through a minimally invasive endovascular approach, with immediate technical and clinical success in less than 24 hours.

Keywords: Pelvic pain, varicose veins, renal veins, embolization, angioplasty.

RESUMO

A dor pélvica crônica é um problema subdiagnosticado e relativamente comum nas mulheres. Alguns autores evidenciaram prevalência de até 15% entre mulheres de 18 a 50 anos, com repercussões sobre a qualidade de vida e sobre a economia. Dentre as causas de dor pélvica crônica, destaca-se a síndrome de congestão venosa pélvica, com quadro clínico caracterizado por diversos graus de dor, disúria, hematúria, dismenorréia, dispareunia e congestão vulvar, que pode ser acompanhado de varizes vulvares, descrito em 1949 por Taylor. Relatamos o caso de uma paciente portadora de dor pélvica crônica, na qual se diagnosticou o pinçamento da veia renal

esquerda entre a aorta e a artéria mesentérica superior, com conseqüente quadro de hipertensão do plexo gonadal esquerdo, varizes pélvicas e sintomas de congestão pélvica. O tratamento realizado constou de embolização das varizes pélvicas, por método minimamente invasivo endovascular, com sucesso técnico e resolução dos sintomas em menos de 24 h.

Palavras-chave: Dor pélvica, varizes, veias renais, embolização, angioplastia.

Introduction

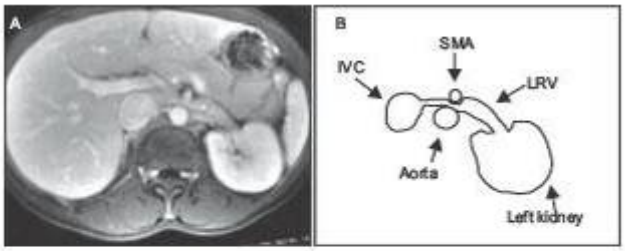
Chronic pelvic pain is a common and underdiagnosed problem in women. In a study including women aged between 18-50 years, Mathias et al. found a 15% prevalence, with major consequences on quality of life and economy.[1] Among the causes of chronic pelvic pain, pelvic congestion syndrome stands out, with clinical status characterized by varied degrees of pain, dysuria, dysmenorrhea, dyspareunia and vulvar congestion, often accompanied by vulvar varices,[2-6] described in 1949 by Taylor.[2]

Laboratory tests for those cases frequently show signs of microhematuria, usually associated with the nutcracker syndrome, an anatomical variation in which the superior mesenteric artery (SMA) and the aorta perform a clamping of the left renal vein, with consequent reflux in the proximal portion of that vein and of the left ovarian vein. The nutcracker syndrome usually affects women aged between 20-40 years, especially multiparous women, and venous reflux causes varicose veins of the deep and superficial pelvic venous plexus, responsible for a clinical status typical of left flank and chronic abdominal pain, besides microhematuria.[3,4,7-11] In men, this syndrome can be manifested similarly, being one of the described causes of varicocele.

Case report

A 35-year-old patient, multiparous, businessperson, sought for clinical care presenting abdominal, pelvic and left flank pain for about 2 years, with daily use of increasing doses of analgesics, including opioids, with no therapeutic success, despite a long investigational history combined with several imaging examinations and laboratory tests.

Physical examination showed flaccid abdomen, painful at palpation of the lower portion and left flank, without signs of peritoneal irritation and absence of vulvar or lower limb varices. Initial laboratory tests revealed microhematuria. In work-up, magnetic nuclear resonance was used, suggesting clamping of the left renal artery between the aorta and the SMA, as well as varices of the deep pelvic venous plexus (Figure 1).



IVC = inferior vena cava; LRV = left renal vein; SMA = superior mesenteric artery.

Figure 1 - Physiopathology: A) tomographic axial section showing clamping of the left renal vein; B) tomography scheme

The patient was referred to the vascular and endovascular surgery service to evaluate possibility of a minimally invasive treatment. She was then submitted to venography, showing clamping of the left renal vein, with significant increase in its proximal diameter, besides extensive varices of the deep pelvic venous plexus, with inverted venous reflux in the left ovarian vein, which also had a diameter about two to three times larger than expected (Figure 2).

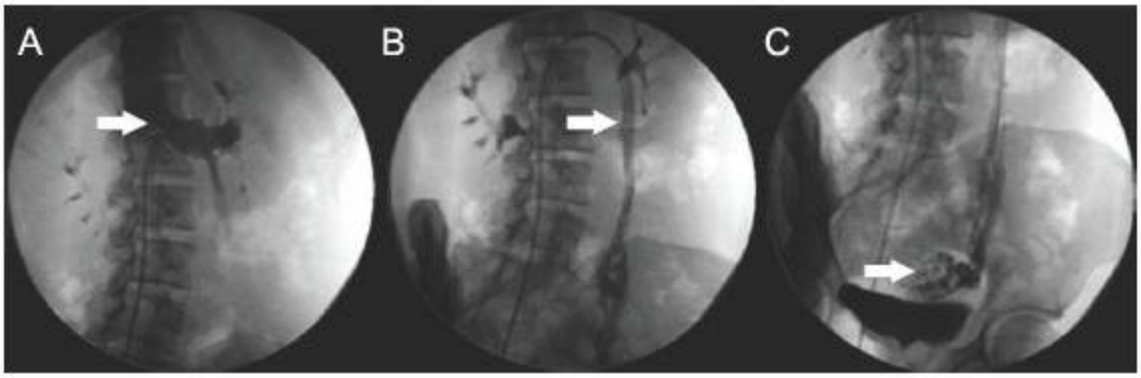


Figure 2 - Diagnostic venographies: A) left renal vein with poststenotic dilatation; B) anomalous reflux in the ovarian vein after contrast injection in the renal vein; C) varices of the ovarian plexus

Two endovascular therapeutic alternatives were initially considered: use of stent in the left renal vein and embolization of pelvic varices, both being procedures reported in the international literature to repair that clinical status.[3,12-18]

Since in many cases there may be therapeutic success only by eliminating pelvic varices, choice was for angioplasty of the left renal vein associated with embolization of the varicosed venous plexus. Through a puncture of the femoral vein, the left renal vein was catheterized and submitted to angioplasty, and the left ovarian vein was embolized using eight Gianturco coils (Cook, USA), with immediate therapeutic success confirmed by almost absence of venous reflux in the affected territory (Figure 3).

Figure 3 - Endovascular treatment: A) balloon angioplasty of the left renal vein; B) varicosed plexus embolized with Gianturco coils; C) angiographic control showing absence of flow in the ovarian varices

Clinical course was satisfactory, with complete regression of pain 24 hours after the procedure. Doppler ultrasound in the immediate postoperative period showed absence of flow in embolized veins, and the patient was discharged 72 hours after the procedure, with suspension of analgesic drugs.

Discussion

Prevalence of the nutcracker syndrome is relatively much higher than its diagnosis, probably because the presence of characteristic anatomic changes does not always cause symptoms.[4]

With regard to cases in which it is confirmedly responsible for symptoms of pelvic venous congestion, lack of clinical reports in the current gynecological and obstetric literature shows that venous congestion is not considered in clinical investigation with adequate frequency. In a study including 66 patients between 1992 and 2000, d'Archambeau et al. reported an 83% incidence of the nutcracker syndrome in patients referred due to symptoms of pelvic venous congestion.[9]

Diagnostic of pelvic venous congestion should be considered among causes of chronic pelvic pain, especially after exclusion of other more common causes, such as pelvic inflammatory disease, endometriosis, interstitial cystitis, pelvic tumors or intestinal inflammatory disease. As to complementary examinations, ultrasound associated with vascular Doppler can be used as a *screening* examination, as long as a proper intestinal preparation is performed and the examiner is experienced enough to identify affected vascular structures. Both magnetic nuclear resonance and tomography can be used, with high sensitivity and specificity, despite being considered invasive.

In d'Archambeau's series,[9] treatment of choice was the endovascular, through embolization of the left ovarian vein and underlying pelvic varicosities, with initial clinical success rate of 86% and reduction of 73% in long-term clinical complaints (mean 43.4 months).

Treatment of pelvic venous congestion and the nutcracker syndrome can be through drug treatment, using estrogens and anti-inflammatory agents, but it has poor therapeutic response.

Surgical treatment, performed through transabdominal or retroperitoneal ligation, or also through laparoscopic approach to the ovarian vein and pelvic varices, has better results, although being more invasive.

Our choice is for the endovascular method, which we believe brings excellent benefits and is the first-choice method in current literature. In addition, it is minimally invasive, as demonstrated in this case, which had excellent clinical course after a 6-month postoperative follow-up.

There is a need of publishing more information on the nutcracker syndrome and on its treatment in varied specialties involved, especially ultrasound, gynecology and urology.

References

1. Mathias SD, Kuppermann M, Liberman RF, Lipschutz RC, Steege JF. Chronic pelvic pain: prevalence, health-related quality of life, and economic correlates. Obstet Gynecol. 1996;87:321-7.

2. Taylor HC Jr. Vascular congestion and hyperemia: the effect on structure and function in the female reproductive system. Am J Obstet Gynecol. 1949;57:211-30.

3. Maleux G, Stockx L, Wilms G, Marchal G. Ovarian vein embolization for the treatment of pelvic congestion syndrome: long-term technical and clinical results. J Vasc Interv Radiol. 2000;11:859-64.

4. Scultetus AH, Villavicencio JL, Gillespie DL. The nutcracker syndrome: its role in the pelvic venous disorders. J Vasc Surg. 2001;34:812-9.

5. Sichlau MJ, Yao JS, Vogelzang RL. Transcatheter embolotherapy for the treatment of pelvic congestion syndrome. Obstet Gynecol. 1994;83(5 Pt 2):892-6.

6. Mathias SD, Kuppermann M, Liberman RF, Lipschutz RC, Steege JF. Chronic pelvic pain: prevalence, health-related quality of life, and economic correlates. Obstet Gynecol. 1996;87:321-7.

7. Ahmed K, Sampath R, Khan MS. Current trends in the diagnosis and management of renal nutcracker syndrome: a review. Eur J Vasc Endovasc Surg. 2006;31:410-6.

8. Hartung O, Grisoli D, Boufi M, et al. Endovascular stenting in the treatment of pelvic vein congestion caused by nutcracker syndrome: lessons learned from the first five cases. J Vasc Surg. 2005;42:275-80.

9. d'Archambeau O, Maes M, De Schepper AM. The pelvic congestion syndrome: role of the "nutcracker phenomenon" and results of endovascular treatment. JBR-BTR. 2004;87:1-8.

10. Itoh S, Yoshida K, Nakamura Y, Mitsuhashi N. Aggravation of the nutcracker syndrome during pregnancy. Obstet Gynecol. 1997;90(4 Pt 2):661-3.

11. Weiner SN, Bernstein RG, Morehouse H, Golden RA. Hematuria secondary to left peripelvic and gonadal vein varices. Urology. 1983;22:81-4.

12. Capasso P, Simons C, Trotteur G, Dondelinger RF, Henroteaux D, Gaspard U. Treatment of symptomatic pelvic varices by ovarian vein embolization. Cardiovasc Intervent Radiol. 1997;20:107-11.

13. Ahmed K, Sampath R, Khan MS. Current trends in the diagnosis and management of renal nutcracker syndrome: a review. Eur J Vasc Endovasc Surg. 2006;31:410-6.

14. Shin JI, Lee JS, Kim MJ. The prevalence, physical characteristics and diagnosis of nutcracker syndrome. Eur J Vasc Endovasc Surg. 2006;32:335-6.

15. Kim SJ, Kim CW, Kim S, et al. Long-term follow-up after endovascular stent placement for treatment of nutcracker syndrome. J Vasc Interv Radiol. 2005;16:428-31.

16. Chen W, Chu J, Yang JY, et al. Endovascular stent placement for the treatment of nutcracker phenomenon in three pediatric patients. J Vasc Interv Radiol. 2005;16:1529-33.

17. Cordts PR, Eclavea A, Buckley PJ, DeMaioribus CA, Cockerill ML, Yeager TD. Pelvic congestion syndrome: early clinical results after transcatheter ovarian vein embolization. J Vasc Surg. 1998;28:862-8.

18. Hartung O, Grisoli D, Boufi M, et al. Endovascular stenting in the treatment of pelvic vein congestion caused by nutcracker syndrome: lessons learned from the first five cases. J Vasc Surg. 2005;42:275-80.

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