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Case Report

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Recurrent Varicocele Associated With Congestion Pelvic Syndrome

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Abstract

Left Renal Vein compression, entitled as Nutcracker Syndrome, usually is described as cause of Pelvic Congestion once the difficult of drainage of the left kidney deviate vein flow to Gonadal vein developing Pelvic varices in women, and Varicocele in men. Recurrence of Varicocele is described to be between 1 to 35%, dependent on the surgical technique used to repair it. Among the cause of recurrent varicocele, include surgical technique failure, low Body Mass Index, venous plexus variation (persistence of branched spermatic veins), and venous compression. In this study, the authors present 2 cases of recurrent varicocele in young men caused by pelvic congestion due to the Left Renal Vein (LRV) and Left Common Iliac Vein (LCIV) compression, treated by endovascular techniques with good results. In the Literature there are few papers relating the association of both syndromes. Authors discuss of the association of recurrent varicocele and pelvic congestion, and also suggest a routine investigation of it in this recurrence.

Abbreviations

NUS: Nutcracker Syndrome
LRV: Left Renal Vein
SMA: Superior Mesenteric Artery
DUS: Duplex Ultrasonography
CT Scan: Computed Tomography Scan
NMR: Nuclear Magnetic Resonance
LCIV: Left Common Iliac Vein (Rotker and Sigman)

Introduction

Nutcracker Syndrome is a condition with a set of symptoms caused by an obstruction of Left Renal Vein (LRV) outflow for the Superior Mesenteric Artery (SMA), known as anterior Nutcracker. When the third portion of duodenum is compressed by SMA causes Wilkie Syndrome El-Sadr and Mina, 1950, first described the phenomenon, but the term "nutcracker was first used [1,2]. Some pathologic conditions that cause compression of the LRV include retroperitoneal tumors, pancreatic neoplasms, SMA aneurysms, among others less common.

Zerhouni et al were the first to report patients presented with varicocele, pelvic pain, and hematuria, and contributed to understand the issues involved in the diagnosis of the nutcracker syndrome in 1979 [2]. They related an elevated left renal vein pressure in three patients with left-sided varicocele and pelvic pain, whereas the right side was normal.

Varicocele affect up to 10% of men, and it is more frequent in the left testicle. Recurrence of Varicocele is described to be between 1 to 35%, dependent on the surgical technique used to repair it. (Rotker and Sigman) The association of recurrent varicocele and venous compression was described to be by 28% in a study of 3041 patients [3].

The aim of this work is to describe two cases of recurrent varicocele associated to Left Renal Vein (LRV) compression, entitled of Nutcracker syndrome that due to recurrence of symptoms were diagnosed and treated successfully.

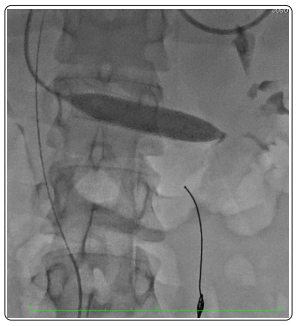
Patient Demographic

Case 1

JVAA, 18 years old, presents with left leg pain and edema, scrotal pain and recurrent varicocele after 2 surgeries, no local trauma or other complaints. After physical examination, it was observed left leg edema. Spermogram count showed 700.000 sperms with 20% of viability. Doppler ultrasound exam showed compression on left renal vein of 53%, and compression on left common iliac vein of 63%. CT scan confirmed left renal vein and left common iliac vein compressions. Bilateral Femoral Phlebography confirmed left renal vein compression with gonadal vein reflux, and left common iliac vein compression with contralateral flow deviation. (Fugure 6,7). The patient was submitted to an Angioplasty with stent first of the Left Common Iliac Vein with good result. In the first treatment both common vein access was used to treat the left common iliac vein compression. An 18x60mm Wallstent (Boston Co) was implanted followed by angioplasty with good result. Left leg edema was solved, but he remained with persistence of scrotal pain. Spermogram count showed 9.000.000 sperms with 82% of viability. So, due to the persistence of scrotal pain he was submitted to an Angioplasty with stent of the Left Renal Vein, and Left Gonadal Vein Embolization with remission of almost all complaints.In this procedure, a right jugular vein access associated with right femoral vein access was used to treat the left renal vein due to the short distance of

Jugular vein to renal vein and to the favorable angle. Left Gonadal vein catheterization was used in order to proceedan embolization using the sandwich technique with foam of polidocanol 1%, and 6 Gianturcocoils of 8,9 and 10mm. After that, a 14x60mm Wallstent (Boston Co) was implanted followed by angioplasty to fix and dilate the stent. In the early Post-operative period the patient observed relieve of testicle pain and pelvic complaints. After 3 months, DUS showed patent stents of left renal vein and left common iliac vein. Last Spermogram count showed 14.000.000 sperms with 92% of viability. Recent DUS (18 months after LRV and 24 months after LCIV treatment) showed patency of both treated veins - Left Renal and Left Common Iliac veins.

Case 1: LRV post stent placement



Case 1: LRV stent placement and angioplasty

Case 2

LSP, 16years old, presented with scrotal pain and recurrent varicocele after 3 surgeries, no local trauma or other complaints. After physical examination, it was observed left leg edema. Spermogram count showed 1.400.000 sperms with 35% of viability. Doppler ultrasound exam showed compression on Left Renal Vein of 65%, and compression on Left Common Iliac Vein of 62%. CT scan confirmed theLeft Renal Vein compression of >60%, and Left Common Iliac Vein compression. Bilateral Femoral Phlebography confirmed LRV compression with Gonadal Vein reflux, and LCIV compression with contralateral flow deviation.First, a right and left common femoral vein access was used to treat the LCIV compression with use of a 16x60mm Wallstent (Boston Co). Subsequently, Right Jugular vein access was used to treat the LRV compression due to a favorable angle between Inferior Cava Vein and Left Renal Vein. After crossing the LRV obstruction a 14x40mm Wallstent was implanted followed by angioplasty with good stent placement. Control angiogram showed no gonadal vein reflux, which canceled the gonadal vein embolization. After 6 months of stent placement, Spermogram count showed 13.000.000 sperms with 95% of viability. DUS of control at 24 months showed both LRV and LCIV stent patency and with good flow.

Results

In case 1, a huge left common iliac vein compression was firstly treated in order to relieve testicle pain, left leg edema, and pain. After some months, he had improvement of leg left edema and pain, also of Spermogram count, but the left testicle pain continued. After some months of clinical treatment without desirable result, he was submitted to the left renal vein treatment, with angioplasty and stent in LRV, and Gonadal Vein embolization with good result in spermatozoid count and testicle pain. In the follow up period, DUS of control showed patency of both vein stents with good stent position and vein flow at 12 and 18 months after the procedure.

In case 2, both vein compression, LRV and LCIV was treated in order to avoid gonadal embolization. First, a right and left common femoral vein access was used to treat the left common iliac vein compression. Right Jugular vein access was used to treat the left renal vein compression due to a favorable angle between inferior cava vein and left renal vein. Control angiogram showed no gonadal vein reflux that canceled the gonadal vein embolization. Patient observed relieve in almost all complaints after the procedure. DUS of 24 months control showed patency of both vein stents with good stent position and vein flow.

Discussion

Nutcracker Syndrome is an anatomical disorder resulted by the compression of Left Renal Vein that results in a renal vein flow deviation to the Gonadal Vein, and Pelvic vein dilation. In the majority of cases, patients complain of left lumbar pain, fatigue, pelvic pain, hematuria, proteinuria, varicocele in men, and pelvic varices in women. The severity of symptoms varies from microhematuria to severe pelvic congestion complaints. They are aggravated by orthostatic position, physical activities, and can evolve to macrohematuria, orthostatic proteinuria, varicocele, testicle pain, and orthostatic intolerance during daily basis activities.

Hematuria is one of the most common symptoms, and can vary from micro to macrohematuria, that can cause huge anemia in some cases. Pain is other frequent symptom commonly referred to left flank that radiates to posteromedial thigh and buttock. It is worsened by sitting and long standing positions, also walking, and riding some vehicles such as bicycles, motorcycles, and horses.

Varicocele affect up to 10% of men, and it is more frequent on the left side. Recurrence of Varicocele is described to be between 1 to 35%, dependent on the surgical technique used to repair it.Recurrence rates also vary dependent on the age of population being studied, indications for the first intervention, types of varicocele, surgical technique employed, definition of the recurrence, and the follow-up period. (Rotker and Sigman) Sen Li et al in a retrospective study of 3042 patients submitted to varicocele surgery repair, 858 was related to presented Left Renal Vein (LRV) compression (28,21%). In the total of patients with recurrent varicocele (18), LRV compression was presented in fifteen (15/18) [4,5].

Jargiello el al in a study of 33 varicocele recurrence found that the majority of patients (66%) had a type III duplication when submitted to phlebography, and that no recurrences were seen after embolization of the Gonadal Vein [6].

In the causes of Varicocele the most important are absence or congenital incompetence of valves in Gonadal vein, or renal vein

obstruction causing flow reversal to Gonadal vein. There are some theories that explain the damage of Varicocele in testicle function; some of them are hyperthermia, hypoxia, hormonal changes, and others.

Duplex ultrasound exam (DUS) is useful to demonstrate vein compression in only 47% of left renal vein, but in 78% in Left Common Iliac Vein (LCIV). DUS criteria for the nutcracker syndrome include the five times increase in flow velocity in the LRV before and in the site of compression is related to have sensitivity of 80% and specificity near to 95% for nutcracker syndrome. Also the post-stenotic peak systolic velocity more than 100cm/s is a good parameter to consider a high-gradecompression [7-9].

Angio CT scan is a good exam that allows the visualization of vein compression and vein dilation, but it depends of the right protocol of the dye injection. Due to the low volume of contrast media injection, and wrong time acquisition sometimes it can causes false negative results. Nuclear Magnetic Resonance (NMR) scan has the same false negative results as Angio CT scan due to use of systemic gadolinium vein injection.

Gold standard exam is still Phlebography that shows the LRV compression, associated with Left Gonadal Vein dilation and reflux. Selective gonadal vein angiography can demonstrategonadal reflux until the left testicle. In both cases related, the diagnosis included DUS, Angio CT scan, and Phlebography to make the right diagnosis of compression and vein reflux.

NMR was not used in these cases

The association of Nutcracker Syndrome and May-Thurner Syndrome is not common. In the majority of cases we observe predominance of the symptoms in only one venous axis. Due that the treatment should be oriented to relieve the main complaints.

Conclusion

The aim of the study was to report the association of both Left Renal and Common Iliac vein compression in the genesis of varicocele recurrence. Both cases were successfully treated using endovascular techniques. The authors also emphasized that are many factors involved in the causes of recurrence, among them the most important seems to be the initial surgical technique used. Nevertheless, compression syndromes have also been described as an important cause of recurrence.

Urologists should pay attention of compression syndromes that can have a special role in the recurrence of varicocele. DUS and Angio CT scan can contribute to clarify such disorders, and these diagnostic tools should be used in order to study the hemodynamic involvements in which contribute to the genesis of varicocele. Despite of not having a consensus of which technique is the best one to treat this occurrence, endovascular treatment seems to be a feasible option [10-18].

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