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Left renal vein transposition using autologous gonadal vein graft in patient with combined anterior nutcracker and May-Thurner syndromes

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SUMMARY

Anterior nutcracker syndrome (ANCS) and May-Thurner syndrome (MTS) are part of a rare group of vascular compression syndromes where extrinsic compression of arteries or veins results in non-specific clinical presentation posing diagnostic and management challenges. We present the case of a female patient in her early 40s with a 3 year history of left flank and pelvic pain, left leg swelling and microscopic haematuria attributed to a rare combination of ANCS and MTS. Compression of the left common iliac vein (MTS) was managed with left common iliac stenting and compression of the left renal vein (LRV; ANCS) was managed surgically with a novel modified technique of LRV transposition using an autologous gonadal vein graft with subsequent resolution of all symptoms. To our knowledge, this is the fourth case presenting the coexistence of the two syndromes in the literature and the first presenting the modified treatment approach.

BACKGROUND

Anterior nutcracker syndrome (ANCS) is characterised by symptomatic compression of the left renal vein (LRV) between the superior mesenteric artery (SMA) and the aorta, resulting in the obstruction of normal blood flow to the inferior vena cava (IVC). Common symptoms are haematuria, proteinuria, persistent left flank pain and pelvic congestion.[1–9] May-Thurner syndrome (MTS) involves the compression of the left common iliac vein (LCIV) between the right common iliac artery (RCIA) and the lumbar spine. Symptoms include swelling of the left lower extremity, varicosities and chronic venous stasis ulcers.[5 8 10] Both syndromes occur predominantly in middle-aged women[2 5 7 8 10] and are otherwise of unknown prevalence.[1 2 6 8] Occurrence of ANCS is thought to correlate with a low body mass index.[3 4 7–9]

Treatment of either syndrome remains a controversial issue and several different options are available, ranging from left renal/gonadal vein transposition or nephrectomy for ANCS,[1 2 4 6] to endovascular stenting for either.[1 2 4 6 11 12] The above controversy is attributed to the rarity of the syndromes and subsequent lack of clinical trials to guide evidence-based practice. We present the fourth case of a patient presenting with both ANCS and MTS ever reported in the literature[7 10 13] and the first reported case managed surgically with a modified LRV transposition technique using an autologous gonadal vein graft,[1–10 13] combined with an endovascular stent placement for MTS. This is with the aim of providing an alternative to the currently used saphenous vein graft for LRV transposition which requires extra dissection, while also providing the added benefit of improving pelvic congestion, likely leading to improved symptom resolution.

CASE PRESENTATION

We present the case of a multiparous female patient in her early 40s with combined ANCS and MTS. She initially presented with chronic pelvic pain, left lower extremity swelling and left flank pain. The patient also complained of a feeling of 'intense swelling' in her pelvic area. The pain had worsened progressively over the last 3 years. She had previously presented to several specialist doctors where she underwent various investigations. This included a gynaecologist, a gastroenterologist and an orthopaedic surgeon, who ruled out any relevant pathologies that could explain her symptoms. Her kidney function was normal, and she had episodes of microscopic haematuria on a midstream urine sample without macroscopic haematuria.

The chronic, intensifying nature of her symptoms and her extensive assessment by other specialists ruled out numerous common differentials. Possible ANCS was also initially detected during an ultrasound which led to her being referred for a CT scan, and to our team for further investigation.

INVESTIGATIONS

Her initial CT angiography demonstrated a reduced angle (32°) between the SMA and aorta (normal range 38–65°), LRV stenosis with associated prestenotic dilatation and significant dilatation of left gonadal vein with formation of pelvic collateral veins (pelvic congestion syndrome) (figure 1). No right gonadal vein dilatation was observed. Given the suspicion of ANCS, the patient was sent for a venography to further investigate the degree of LRV stenosis.

Selective catheterisation of the LRV distal to the origin of the left gonadal vein demonstrated reflux of contrast in the left gonadal vein and catheterisation of the proximal left gonadal vein demonstrated stasis with no evidence of antegrade flow to the LRV (Figure 2A). In addition, significant LCIV stenosis was observed, secondary to compression by the RCIA at the

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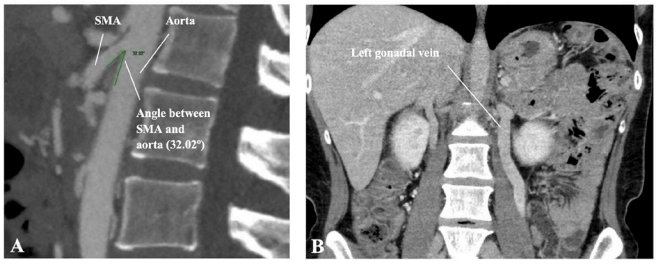

Figure 1 (A) CT image showing the angle between the aorta and SMA, measuring 32.02° preoperatively. (B) CT image showing congestion and dilatation of the left gonadal vein preoperatively. SMA, superior mesenteric artery.

level of crossing with the RCIA with a significant decrease of the intraluminal area at the compression site. Partial drainage of the left gonadal vein was demonstrated to the left internal iliac vein. The above confirmed ANCS and added a diagnosis of MTS.

TREATMENT

MTS was treated first, given the less invasive treatment and the possibility that there would be significant enough improvement to the patient's symptoms that more complex intervention for ANCS could be avoided. This was done endovascularly via deployment of a self-expanding uncovered 16 × 80 mm (abre venous stent—Medtronic) in the left common iliac vein following confirmation of a haemodynamically significant extrinsic compression of the left common iliac vein from the RCIA by intravascular ultrasound and venography.

The procedure was successful and patency of the left common iliac vein endoprosthesis was confirmed on US Doppler, follow-up venography (figure 2B) and MR angiography. There was improvement in lower extremity swelling and partial pelvic pain improvement as initially expected, but the left flank pain persisted, something we attributed to the coexistence of ANCS. The decision to get a follow-up venography was made to reassess the degree of LRV stenosis and the extent of pelvic congestion before considering further treatment for ANCS.

Follow-up venography and selective catheterisation of the LRV distal to left gonadal vein origin demonstrated reflux of contrast in the left gonadal vein. Subsequent selective catheterisation of left gonadal vein demonstrated stasis proximally

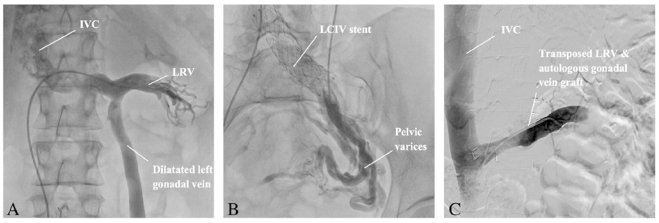

Figure 2 (A) Venography image showing compression of the LRV proximal to the origin of the gonadal vein with reduction of contrast flow to the IVC and flow of contrast to the dilatated left gonadal vein. (B) Venography image showing the formation of pelvic varices, post LCIV stent placement. (C) Venography image showing restoration of contrast flow in the transposed left renal vein to the IVC postoperatively. IVC, inferior vena cava; LCIV, left common iliac vein; LRV, left renal vein.

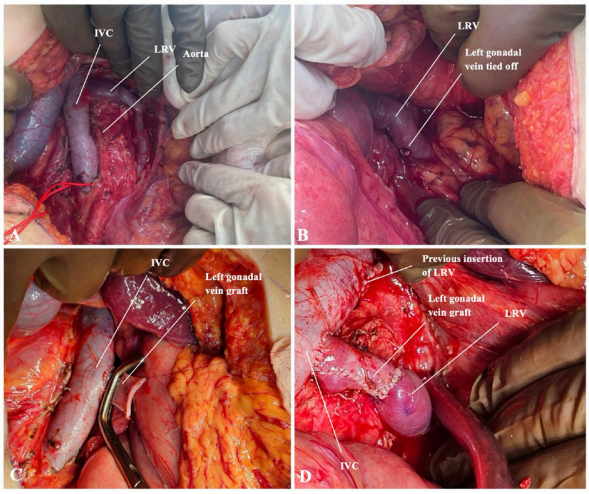

Figure 3 (A) Intraoperative image showing the dilatated LRV up to the level of imprint of the SMA. (B) Intraoperative image showing the dissection of the left gonadal vein from the LRV. (C) Intraoperative image showing the anastomosis of the restructured left gonadal vein graft. (D) Intraoperative image showing end-to-side anastomosis of the left gonadal vein graft to the IVC and LRV with normal blood flow. Note the previous insertion of the LRV on the IVC cephalad to the anastomosis. IVC, inferior vena cava; LRV, left renal vein; SMA, superior mesenteric artery.

and reflux with pelvic congestion distally confirming haemodynamically significant compression of LVR in keeping with ANCS. The decision to intervene for ANCS was made, and follow-up CT imaging was obtained to guide our decision-making as to which approach would be more beneficial to the patient.

The follow-up cross-sectional imaging confirmed ongoing congestion in the patient's pelvic region, with a large network of veins arising from a dilatated left gonadal vein. The great degree of congestion excluded the option of autotransplantation, given the possibility of symptom continuation or exacerbation. Hence, the remaining available options were either endovascular stenting, nephrectomy or LRV transposition.

Endovascular stenting was ruled out given the high complication risk. The patient was given the option of altruistic donation or LRV transposition, of which she chose the latter. The choice to use the gonadal vein as a graft was proposed both to lengthen the LRV, allowing it to be transposed to the IVC with greater ease, and to assist in relieving the great pelvic congestion which we pinpointed as the cause of the majority of the symptoms.

Midline laparotomy was performed, followed by Cattell-Braasch and Matox manoeuvres. Kocherisation of duodenum was performed, with exposure of the entire length of the LRV (figure 3A). This was followed by identification, dissection and division of the insertion of the left adrenal and left gonadal veins to the LRV (figure 3B). A large section of the left gonadal vein, approximately 4 cm in length was dissected and prepared for the elongation of the LRV using 6.0 continuous proline suture (figure 4). The autologous graft was first anastomosed to the medial wall of the IVC around 4 cm caudally from the insertion of the LRV (figure 3C), using a Satinsky clamp to partially occlude the IVC. Once the anastomosis was complete, the Satinsky clamp was applied to the graft itself to release the flow of blood in the IVC. Next, the LRV was cut at its origin to the IVC, the orifice was sutured,

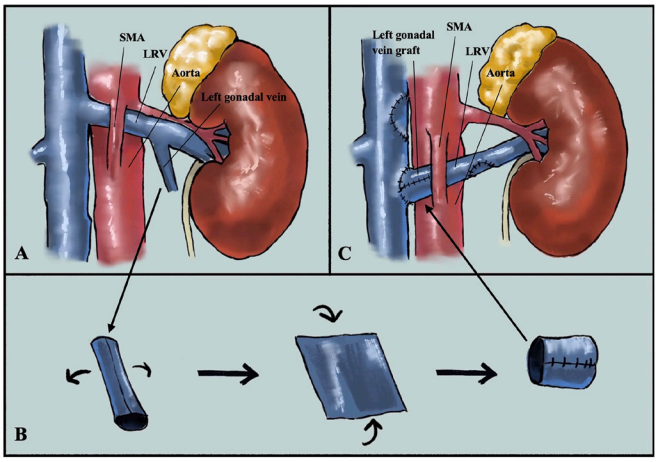

Figure 4 (A) Diagrammatic representation of the anatomy of anterior nutcracker syndrome, with the LRV compressed between the SMA and aorta. (B) Diagrammatic representation of the use of the left gonadal vein as an autologous graft for elongation of the renal vein. An adequate length of gonadal vein (at least the circumference of the renal vein) is dissected away and opened longitudinally. The lateral ends are then stitched together to create the graft. (C) Diagrammatic representation of the post-transposition anatomy, showing the anastomosis of the LRV to the inferior vena cava caudal to the original position, using the graft. LRV, left renal vein; SMA, superior mesenteric artery.

and the renal vein was transposed to the open end of the autologous graft, taking care to avoid twisting or kinking of the vein (figure 3D). Estimated blood loss was 30 mL and there was no requirement for transfusion.

OUTCOME AND FOLLOW-UP

After both operations were completed, there was an immediate and significant improvement in the patient's symptoms and quality of life. Pelvic pain and left flank pain were both resolved, as well as her feeling of pelvic congestion. Patency of the LRV transposition was confirmed on imaging on a 4 month follow-up with CT angiography and venography. Normal antegrade flow of contrast from the LRV to the IVC was noted (figure 2C), as well as the absence of renal vein collaterals. The patient remained on clopidogrel (75 mg once per day) due to her LCIV stent.

DISCUSSION

Diagnosis of MTS involves CT/MRI or MR venography in correlation with the patient's symptoms and is established intraoperatively using intravascular ultrasound (IVUS). Treatment usually follows presentation with DVT (which, however, was not seen in our patient) and often involves thrombolysis followed by placement of an endovascular stent[8 11 12] in the LCIV, with RCIA repositioning also rarely seen.[8]

Moudgil *et al*, in a literature review, reported excellent results for the endovascular treatment of MTS, with a primary patency of around 95% in the studies they reviewed, and a patency of 90–100% at 1 year post-stent placement.[11]

With regard to ANCS, in addition to the clinical features of left flank pain, pelvic pain and haematuria,[1–9] imaging features on CT/MR include reduced aortic-SMA angle (<38°, although some suggest larger or smaller angles),[2 4 5 9] LRV stenosis and a dilatated left gonadal vein as the main collateral pathway. On conventional renal venography, a pressure gradient of >3 mm Hg is observed in early ANCS.[2 5–7]

Treatment options for ANCS remain controversial, and there is still no standardised approach, possibly due to its rarity. Conservative management is initially preferred, especially in paediatric and adolescent patients, due to the high likelihood of spontaneous resolution of symptoms with age.[1–3 7 9]

Endovascular treatment (stenting) or surgical options should be carefully selected and tailored according to the individual patient. Although endovascular stenting of the LRV provides a less invasive approach, it is not without significant complications. Stent occlusion is seen to occur in some cases,[2–4 6 7] which is likely to require reoperation. In addition, there is a risk of stent migration (the literature suggests between 7 and 9%,[2 3] although a study by Korkes *et al* claims it is under-reported and could be as high as 20%[3]), which could pose a life-threatening situation for the patient, for example, if the stent migrates to the heart, requiring major operations and a long recovery.

Few studies on endovascular stenting for ANCS exist other than case reports and case series. Hartung *et al* published a series of five patients treated for ANCS through stenting, showing short-term improvement in all five patients. The study also shows some of the difficulties associated with the procedure, given it reported two stent migrations at 4 months postoperatively, which was attributed to the use of shorter stents (40 mm).[13]

LRV transposition is the most common surgical approach used to treat ANCS with various techniques described in the past, including the use of synthetic, allogenic or autologous grafts.[1 2 4 6] Other surgical options, including autotransplantation, renocaval bypass and SMA transposition,[2 5 7 8] are used less often than renal vein transposition mainly due to their complexity and risk of complications. Left nephrectomy could also be performed as a treatment of refractory ANCS but should be reserved as the last option due to its radicality and the loss of an otherwise fully functional kidney. In those cases, altruistic kidney donation to a patient in need of renal transplant should also be considered.

Reed *et al* reported the results of 11 transposition operations in 2009 with excellent results. Of those patients, 27% (3/11) required reintervention postoperatively, and of those three, two had been found to have thrombosed LRVs intraoperatively, which then developed into postoperative LRV thrombosis. One patient required stenting, while the others underwent transposition of the left gonadal vein into the IVC.[4]

In our case, the chronic nature and persistent intensifying symptoms of the patient did not support the continuation of conservative treatments. Given the less invasive approach used, we chose to proceed with the treatment of MTS first. After proceeding with the endovascular stenting of the LCIV, the patient was assessed 1 month postoperatively, where no resolution of symptoms of left flank and pelvic pain was observed.

The operation for ANCS was performed nearly 1 year post-LCIV stenting, which required us to repeat a CT scan and venography in the planning of intervention. We chose not to proceed with an endovascular approach due to the high risk of stent migration and thrombosis. Additionally, we recommend that in cases with combined MTS and ANCS, autotransplantation should be avoided, given the kidney would be re-implanted to an area of pre-existing congestion, risking the continuation of symptoms.

The use of an autologous gonadal vein graft for LRV transposition is a novel technique that has previously been used for the extension of the renal vein in kidney transplantation.[14 15] The convenience of using the gonadal vein as a graft in LRV

transposition arises from its natural dilatation resulting from ANCS, as well as the ability to harvest it through the same incision, not requiring further dissection. This makes it preferable to a saphenous vein graft.

This natural dilatation of the gonadal vein has already been used in gonadal vein transposition for ANCS; however, symptom relief is variable. Gilmore *et al*, in a case series, reported complete symptom relief in 61.1% of patients undergoing gonadal vein transposition.[16] This shows disconnecting the gonadal vein from the collaterals alone is not enough for many patients to achieve full recovery, also confirmed by Reed *et al*, who report that many of the symptoms of ANCS are related to LRV compression and resolve after LRV transposition.[4] Our approach aims to relieve LRV compression, simultaneously ligate the gonadal vein and use it as an autologous graft to extend the LRV and prevent stretching. It is, therefore, likely to achieve the best symptom relief. This also makes it ideal in cases with coexisting ANCS and MTS, as both cause pelvic congestion,[8] which likely has a compounding effect when seen together.

Although endovascular treatment with combined LRV stenting and gonadal vein ligation/embolisation shows success in relieving ANCS symptoms,[17] it is not without significant complications, as described above. Our technique uses the same principle (simultaneous relief of LRV compression and disconnecting the gonadal vein from the collaterals) without the risks of stent migration or the need for coagulation postoperatively, possibly making it preferable to endovascular approaches.

It should be noted that some authors have reported thrombosis after LRV transposition.[4 18] Although we cannot definitively rule this out in our approach, we note that our use of an autograft to lengthen the LRV may decrease the risk of thrombosis, given it reduces the risk of vascular stretch injury.[19] Nevertheless, alternatives to transposition should be considered in high-risk individuals.

In conclusion, the use of an autologous gonadal vein graft can provide a safe alternative to the saphenous vein graft or allograft for LRV transposition in ANCS and combined ANCS and MTS with several positives. It can also help relieve pelvic congestion by disconnecting the LRV from the pelvic collaterals, assisting in the resolution of associated symptoms while removing the need for anticoagulant therapy or the risks of stent occlusion and migration.

Patient's perspective

I have been suffering from left-side back pain and pelvic pain for many years. It was not easy at all to make the diagnosis and understand what was causing my symptoms. I am very thankful to get the correct diagnosis and the appropriate treatment. I realised from the beginning that what I had was extremely rare and that the treatment may not resolve all my symptoms. It took a long time between the endovascular procedure and the surgery that followed but it was worth waiting. I am very happy since my quality of life has improved significantly and the pain I had on a daily basis has disappeared.

Learning points

- Anterior nutcracker syndrome and May-Thurner syndrome (MTS) can, in rare cases, present together.
- The use of an autologous gonadal vein graft in left renal vein (LVR) transposition can provide a safe and effective alternative to the use of a saphenous vein graft or the use of an allograft.
- In cases with severe pelvic congestion, severing of communication between the LVR and pelvic region (through the ligation and use of the gonadal vein as an extension graft) may assist in the resolution of related symptoms.
- In cases of combined anterior nutcracker syndrome and MTS, autotransplantation is contraindicated due to extensive pelvic collaterals and congestion.

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