

Pelvic congestion syndrome: increasing awareness is the key to facilitating proper diagnosis and appropriate treatment

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Abstract

Pelvic Congestion Syndrome (PCS) represents a complex and frequently misdiagnosed disease that may greatly impact on quality of life of affected patients. An understanding of its complex nature is crucial for both an accurate diagnosis and effective management. In the present paper, a comprehensive overview of the clinical presentation, diagnostic strategies, and the latest advance-

ments in the treatment of PCS is provided. Our aim is to raise awareness of treating physicians, improve proper diagnosis, and promote appropriate treatment.

Introduction

Pelvic Congestion Syndrome (PCS), also known as pelvic venous insufficiency or pelvic venous congestion, is an uncommon but disabling disease that was first described in 1857 by Richet as a condition related to the chronic dilatation of pelvic veins.¹ In 2009, PCS was defined in the VEIN-TERM transatlantic interdisciplinary consensus document as a syndrome characterized by “chronic symptoms, which may include pelvic pain, perineal heaviness, urgency of micturition and postcoital pain, caused by ovarian and/or pelvic veins reflux and/or obstruction, and which may be associated with vulvar, perineal, and/or lower extremity varices”.²

The real prevalence of PCS is unknown because the diagnosis is often very challenging due to the wide range of clinical presentations that may frequently mimic other gynecological or gastrointestinal disorders.² Hence, patients affected with PCS, who are predominantly women, experience a significant deterioration in their quality of life, and they frequently attend several consultations with various specialists before receiving a proper diagnosis.¹ This time-consuming process negatively impacts their physical and psychological well-being, so the disease is often accompanied by depression (2.5-50% of cases), anxiety (10-20% of cases), and somatic complaints (10-20% of cases). Hence, the aim of treatment is not only to alleviate symptoms but also to improve the quality of life.³

Clinically, patients affected with PCS most commonly report pelvic pain and discomfort, often exacerbated by prolonged standing or sitting and involving the abdomen, pelvis, and legs.¹ PCS deeply impacts the quality of life, but most cases remain untreated. A multidisciplinary approach to this condition is crucial to allow prompt and accurate diagnosis.

Increased awareness among treating physicians about PCS is essential to facilitate proper identification and appropriate treatment. Hence, in the present paper, we aim to provide a comprehensive overview of PCS, focusing on clinical presentation, diagnostic strategies, and the latest advancements in treatment.

Epidemiology, etiology, and pathophysiology

The prevalence of PCS is challenging to ascertain as it is frequently underdiagnosed or misdiagnosed. The male-female ratio seems to be 1:30,⁴ as hormones may impact the development of the disease that is demonstrated to be less symptomatic following menopause. Recent studies suggest that PCS affects approximately 10-15% of women of reproductive age, with a higher incidence in multiparous women and those with a history of pelvic inflam-

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Key words: Pelvic Congestion Syndrome, chronic pelvic pain, venous insufficiency, pelvic varices, transcatheter embolization.

Contributions: conception and design, GMP, OB; analysis and interpretation, GMP; data collection, GMP; writing the article, GMP, OB; critical revision of the article, OB, YT. All authors have read the published version of the manuscript, and agreed to be held accountable for all aspects of the work.

Conflict of interest: the authors declare no potential conflict of interest.

Funding: none.

Ethics approval and consent to participate: not applicable.

Informed consent: not applicable.

Availability of data and materials: all data generated or analysed during this study are included in this published article.

Received: 31 July 2024.

Accepted: 12 September 2024.

Early view: 11 November 2024.

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Veins and Lymphatics 2024; 13:12883

doi:10.4081/vl.2024.12883

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matory disease or pelvic trauma. In patients suffering from chronic pelvic pain, the prevalence of the disease is up to 30%.⁵

The etiology of PCS remains poorly understood but derives from Pelvic Vein Insufficiency (PVI), although it has been calculated that only 60% of patients with PVI finally develop PCS.⁵ PVI is the incompetence of the internal iliac veins and the inferior vena cava, through the ovarian veins. Internal iliac veins and ovarian veins indeed represent a complex and interconnected network: the ovarian plexus drains into the ovarian veins, while the hemorrhoidal, utero-ovarian, sacral, and vesicular venous plexuses drain into the internal iliac veins.

Several factors contribute to its development, including genetic predisposition, anatomical abnormalities, hormonal influences (estrogens have vasodilatory effects that may exacerbate venous dilation), venous hypertension, and valvular incompetence within the pelvic venous system.^{1,3} Additionally, an increased intra-abdominal pressure during pregnancy, especially in multiparous women, or obesity may aggravate this condition, leading to retrograde blood flow, persistent vascular changes, and finally, the development of pelvic varices (Table 1).

PVI may occur in the absence or incompetence of venous valves (primary PVI). Historical data shows that the congenital absence of ovarian vein valves may occur in 6% of the patients on the right side and between 13% to 15% on the left side, while valve incompetence seems to occur in 35-46% of women on the right and 41-43% on the left side.⁶

Secondary PVI conversely results from external compression hindering the venous outflow. Underlining causes include the “nutcracker” phenomenon (compression of the left renal vein between the superior mesenteric artery and the abdominal aorta), the May-Turner syndrome (compression of the left common iliac vein by the right internal iliac artery), a left renal vein or inferior vena cava tumoral thrombosis, cirrhosis, and a retro-aortic left renal vein.^{3,6}

Clinical presentation and diagnosis

The main symptom (occurring in up to 30% of patients)⁷ of PCS is Chronic Pelvic Pain (CPP). This is a persistent or intermittent pain that gets worse after prolonged standing, sitting, or during menstruation and that lasts for more than 6 months. Pre-treatment pain level measured with Visual Analogue Scale (VAS) is reported to be around 7.34 ± 0.7 , while decreases 0.78 ± 1.2 following interventional treatment.⁸

CPP is typically described as dull, aching, or pressure-like pain and may be associated with dyspareunia, dysmenorrhea, and urinary frequency or urgency. Due to its non-specific symptoms,

patients frequently see many specialists without receiving proper diagnosis or treatment, making this condition a significant economic burden on health budgets. Indeed, according to Riding *et al.*, CPP is responsible for 20-30% of gynecological outpatient appointments in the United Kingdom and has an annual pan-European economic cost of € 3.8 billion.⁹

CPP may occur either alone or in combination with vulvar varicosities and/or lower extremities venous insufficiency. Imaging should thus be used to exclude other causes of CPP as well as to confirm the clinical suspicion of PCS.¹⁰ When ultrasound is used, the diagnostic criteria previously described by Park and colleagues¹¹ should be applied. PCS is hence diagnosed if the exam shows dilated (>4 mm) ovarian veins, tortuous pelvic veins with a diameter >6 mm, dilated tortuous arcuate veins in the myometrium that communicate with bilateral pelvic varicose veins, slow blood flow (less than three cm/s), and reversed caudal or retrograde venous blood flow, particularly in the left ovarian vein or polycystic changes in the ovaries.¹²

The Magnetic Resonance Imaging (MRI) or Computed Tomography (CT) may provide further anatomical details. MRI should be preferred as prevents the exposure to high radiation dose in this population of normally young patients. Computed angiography may show pelvic tortuous para-uterine varices extending laterally in the broad ligament and reaching the pelvic wall or extending inferiorly to communicate with the paravaginal venous plexus. The exam is also pivotal to definitively identify the Nutcracker and May-Turner syndromes.^{12,13}

Venography represents the gold standard, allowing us to visualize venous reflux and varices through contrast dye injection into the pelvic veins.¹⁴ A previous prospective study by Ascuitto and colleagues has shown that MRI has concordance with phlebography in 96% of cases for venous anatomy and 70% for the grade of venous congestion with a sensitivity and specificity of 88% and 67% for ovarian veins, 100% and 38% for internal iliac veins and 91% and 42% for the pelvic plexus respectively. More recent MRI acquisitions (time-resolved or velocity-encoded gradient-echo MRI) are even more accurate, with a specificity, sensitivity, and accuracy of about 61-75%, 100%, and 79-84%, respectively.¹⁵⁻¹⁷

Treatment options

Given the multifactorial nature of PCS, a multidisciplinary approach involving different specialists (including interventional radiologists, gynecologists, neurologists, urologists, gastroenterologists, pain specialists, physical therapists, and psychotherapists) is essential for the comprehensive evaluation and management of affected patients.

Table 1. Risk factors for Pelvic Congestion Syndrome (PCS).

Risk factors	Description
Gender	Women are more prone to pelvic congestion syndrome due to anatomical and hormonal differences (male-female ratio 1:303)
Age	Women of reproductive age, particularly between 20 and 45 years old, are at higher risk
Multiple pregnancies	Pelvic veins increase in size during each pregnancy and may not return to their normal size
Hormones	High levels of oestrogen may contribute to vein dilation
Vascular anomalies	Congenital malformations of the pelvic veins
Family history	Family history of varicose veins or other vascular conditions are associated with increased risk of PVI
Obesity	Overweight may increase pressure on pelvic veins
Sedentary lifestyle	Lack of physical exercise may contribute to the weakness of venous walls
Jobs	Jobs requiring long periods of standing or sitting and that do not allow proper blood circulation may increase the risk of PVI

PVI, Pelvic Vein Insufficiency.

Temporary relief may be achieved with conservative management, including pelvic floor physical therapy, which relies on exercises that strengthen the pelvic floor muscles and improve pelvic alignment, and medical therapy (including analgesics, Non-Steroidal Anti-Inflammatory Drugs (NSAIDs), Gonadotropin-Releasing Hormone (GnRH) antagonists with Hormone Replacement Therapy (HRT), dihydroergotamine and medroxyprogesterone acetate). Long-term pharmacological therapy is not recommended for the treatment of PCS because of the possible adverse effects and the limited efficacy.¹⁸

Psychotropic drugs have proven effective in treating chronic pelvic pain. The most used are Gabapentin (GBP) and Amitriptyline (AMI) (Table 2).¹⁹ Oestrogen may have vasodilatory effects, suggesting that a hypoestrogenic state could potentially improve symptoms. However, definitive conclusions cannot be drawn as the data currently at our disposal are poor and reported in only small studies. Patients with refractory symptoms may require interventional strategies.⁵

Surgical options include laparoscopic ovarian veins ligation or stripping, abdominal hysterectomy, and oophorectomy. Hysterectomy, with or without bilateral salpingo-oophorectomy, is not widely adopted, while transcatheter pelvic veins embolization is now considered the gold standard and recommended by the Society of Vascular Surgery with a 2B level of evidence.²⁰

The procedure is performed using catheters inserted through venous access (more often the basilic vein) into the abnormal pelvic veins under fluoroscopic guidance. A second access through the right common femoral vein may be used to improve visualization of the confluence of the inferior vena cava. Firstly, diagnostic venography is done through a multipurpose catheter; then, the gonadal and internal iliac veins are selectively cannulated. The contrast medium is injected into the vessel of interest, and the patient is asked to perform a Valsalva manoeuvre so the physician can assess for reflux. Retrograde flow toward the pelvis is diagnostic of reflux. Vessels showing a significant increase in caliber and retrograde venous flow are directly embolized using the catheter already in place. In men, embolization techniques are applied in the treatment of varicocele, which causes symptoms similar to PCS to occlude the left spermatic vein and, less frequently, the right one, the vas deferens, and the cremasteric vein.^{21,22}

There is no definitive evidence on the optimal material, but several embolic agents have been described in the literature, including sodium morrhuate, Sodium Tetradecyl Sulphate (STS), n-butyl-2-cyanoacrylate (glue), ethylene-vinyl-alcohol copolymer, and lauromacrogol 400 used in combination or not with occlusive detachable plugs and coils, such as platinum coils (MRI-compatible up to 1.5 Tesla).¹⁴

Anyhow, the Tessari technique is the most employed and involves using 3-6 mL of 3% Sodium Tetradecyl Sulphate (STS) mixed with air or gel foam. This foam is injected into the dysfunctional veins below the pelvic brim, optionally combined with balloon occlusion to aid stagnation or followed by embolization with coils approximately 40-50 mm long (the “sandwich” technique) to cover up to 5 cm from the origin of the gonadal veins, the left renal vein, or the inferior vena cava, taking care to close any collateral branch to avoid recurrences.

In select cases, when pelvic vein stenosis or obstruction exists, endovascular angioplasty and stenting may be considered to restore venous patency and treat the compression. Indeed, preliminary data suggest that treating the stenosis may be more crucial for relieving pelvic pain than addressing concurrent reflux in the gonadal veins, if both issues are present.²³

Stents used in the venous system are generally self-expandable with diameters ranging from 16 to 20 mm. In some selected cases in which a high radial strength is required a balloon-expandable stent may be needed.

Recently, dedicated self-expanding venous stents have been developed, including the VICI VENOUS STENT®, Veniti, SINUS-VENOUS STENT, and ZILVER 635® despite the current literature still lacking definitive data about their performance.

Clinical outcomes and complications

The previously mentioned studies demonstrated that the medical management of PCS may be beneficial in the short term while the long-term effectiveness of such strategy in controlling the symptoms is unproven.

With that regard, Nicholson and colleagues demonstrated that GBP alone or in combination with AMI is more effective than AMI alone in the treatment of female chronic pelvic pain. Indeed, after 6, 12, and 24 months, patients receiving GBP either alone or in combination with AMI reported a significant improvement in pain when compared to those receiving AMI alone.¹⁹ MPA has proven to relieve symptoms in about 40% of patients, while combining MPA with psychotherapy may be effective in approximately 60% of patients. Anyhow, some patients do not tolerate MPA side effects such as weight gain and depression.²⁴ Indeed, GnRH agonists have several limitations, including side effects, high costs, and unsuitability for extended use due to the risk of menopausal symptoms and osteoporosis. Long-term use of progestins appears to be more beneficial than GnRH analogs, with nearly 80% of women reporting satisfaction 3 months after starting treatment.²⁴

In a prospective open-label study in which 23 women suffering from CPP were randomly assigned to a subcutaneous insertion of

Table 2. Treatment strategies and related outcomes.

Treatment strategy	Description	Pain relief	Author, year
Physical therapy	Exercises to strengthen pelvic floor muscles and improve pelvic alignment	up to 23% ³⁹	Leung-Wright <i>et al.</i> 2020
Psychotherapy	In association with MPA	60%	Schindler <i>et al.</i> 1989 ³⁷
Medical management	Include (NSAIDs), (GnRH) antagonists with (HRT), AMI, GBP, dihydroergotamine, progestins and MPA	40% with MPA 80% with Implanon® 95% with GBP	Schindler <i>et al.</i> 1989 ³⁷ Gezginc <i>et al.</i> 2007 ²⁸ Shokeir <i>et al.</i> 2009 ²⁹
Endovascular management	Pelvic and ovarian vein embolization	88.9% of women had immediate pain relief	Cordts <i>et al.</i> 1999 ³³
Laparoscopic surgery	Trans peritoneal ovarian vein ligation	75%	Gargiulo <i>et al.</i> 2003 ³⁰
Open surgery	Hysterectomy	66%	Beard <i>et al.</i> 1991 ²⁰

NSAIDs, Non-Steroidal Anti-Inflammatory Drugs; GnRh, Gonadotropin-Releasing Hormone; HRT, Hormone Replacement Therapy; AMI, Amitriptyline; GBP, Gabapentin.

Implanon® (a progestin drug that suppresses follicular development and steroid production and induces a state of hypoestrogenism), Shokeir *et al.* described an improvement in symptoms during 12 months in the treated group demonstrating that this drug may represent a valuable and effective hormonal therapy for the long-term treatment of properly selected patients with pelvic pain exclusively related to PCS.²⁵

Surgery is now rarely performed because it is more invasive than endovascular embolization procedures, requires general anesthesia, and has a longer recovery period. Surgical treatment options include laparoscopic ovarian vein ligation (with a reported success rate of up to 100% of cases),²⁶ abdominal hysterectomy, and oophorectomy that may be offered when all other treatments have failed but may not be curative in all cases (22-33% of women still suffer from pelvic pain after treatment).^{14,27}

Comparative studies are lacking, but pelvic venous embolization is currently considered the best approach in the treatment of PCS.⁴

In the study conducted by Kim *et al.*, a total of 127 patients were analysed, of whom 106 received bilateral ovarian veins sclerotherapy followed by coil embolization, and 21 underwent unilateral embolization and sclerotherapy.²⁸ The results indicated that 83% of patients experienced symptom relief, 13% reported no improvement, and 4% reported worsening of symptoms. Accordingly, in a series of 9 patients, Cordts and colleagues reported that about 88.9% of women had immediate relief of symptoms following coil embolization of the ovarian veins and internal iliac tributaries, while the individual symptoms relief varied from 40-100% during the follow-up period (mean follow-up of 13.4 months).²⁹

Data currently available in the literature show that this approach is not linked with major complications, guarantees a technical success rate of 96-100%, and symptoms relief in 75% of patients (with up to 60% experiencing complete resolution of symptoms), but recurrence may happen in 8% of cases.³⁰⁻³³

After the embolization procedure, patients may experience “post-embolization syndrome,” characterized by mild to moderate pelvic pain and fever, which lasts a few days and can be managed with analgesics. Other extremely rare complications described in the literature include coil migration to the pulmonary artery or the left renal vein.³¹

The potential impact on future fertility and the reproductive system in general is one of the main concerns associated with this treatment. A small cohort study on 12 PCS female patients reported that 66.7% of them got pregnant following the procedure, with 6 cases progressing to a live birth, and no differences in pre- and three months post-embolization level of LH and FSH ($p < 0.05$) were found.³⁴

Future perspectives

Further education among specialists in different fields will allow in the future an easier recognition of PCS, and the cooperation of a multidisciplinary team, including gynecologists, vascular surgeons, physiotherapists, psychologists, and primary care physicians, will provide better long-term results rather than just acute management. Hence, further future research is needed to refine international guidelines with the aim to establish specific protocols for definitive PCS treatment.

Conclusions

PCS represents a challenging condition both in terms of diag-

nosis and management. Further efforts are needed to better understand and improve the awareness of this condition among physicians from different subspecialties.

A multidisciplinary approach involving vascular surgeon, gynecologists, interventional radiologists, and pain specialists is essential for the accurate diagnosis, and the application of tailored treatment strategies with the aim to optimize the patient's clinical outcomes and quality of life.

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