Management of Patients when Superficial Venous Disease Arises from Pelvic Escape Points

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Abstract

Chronic pelvic pain (CPP) is a common condition in women that carries with it significant morbidity. It is commonly seen in patients presenting to obstetrics and gynecology outpatient clinic visits. CPP is a presenting symptom of various pathologies including pelvic varicocele, pelvic adhesions, spastic colon syndrome, uterine fibroids, endometriosis, and psychosomatic disorders. Pelvic congestion syndrome has more recently been termed "pelvic venous insufficiency (PVI)" due to the underlying retrograde flow through incompetent ovarian and pelvic veins that are thought to cause the symptoms of CPP. Pelvic varices can commonly present alongside vulvar, perineal, and lower extremity varices. There are some predictable "escape pathways" for these varices that may present for interventional treatment. This article introduces the reader to current terminology, clinical presentation, diagnosis, and treatment of patients with pelvic varices due to PVI.

Keywords
- pelvic varices
- pelvic congestion syndrome
- pelvic venous insufficiency
- embolization
- sclerotherapy
- interventional radiology

Chronic pelvic pain (CPP) is a common condition in women with prevalence varying from 5.6 to 30.9%.[1,2] It results in disruption of daily activities, adversely affects the quality of life, and is the primary presenting symptom in 10 to 40% of all obstetrics and gynecology outpatient clinic visits.[3,4] CPP is a presenting symptom of various pathologies including pelvic varicocele, pelvic adhesions, spastic colon syndrome, uterine fibroids, endometriosis, and psychosomatic disorders.[5–7]

Pelvic congestion syndrome (PCS) has more recently been termed "pelvic venous insufficiency (PVI) due to the underlying retrograde flow through incompetent ovarian and pelvic veins that are thought to cause the symptoms of CPP.[8] PVI occurs in 10% of the female population with at least 30% who have CPP.[9] Patients with PVI can develop pelvic varices which appear on ultrasonography as dilated and tortuous veins in the parauterine or paraovarian areas.[10] Pelvic varices can commonly present alongside vulvar, perineal, and lower extremity varices.

Terminology

Chronic Pelvic Pain

CPP is the intermittent or constant pain in the lower abdomen or pelvis of a woman occurring for at least 6 months in duration, not occurring exclusively during menstruation or intercourse, and not associated with pregnancy. This pain can have a significant impact on a woman's functional ability.[11]

Pelvic Venous Disorders

Pelvic venous disorders in women, also known as PCS or pelvic venous incompetence, is a condition in some women that manifests as chronic, dull pelvic pain in the lower abdomen (more than 6 months of evolution), which can

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intensify with the Valsalva maneuver and decrease or even disappear in the prone or decubitus position.[12]

Pelvic Venous Insufficiency

PVI is the underlying pathology behind the development of pelvic varices. It most commonly occurs due to valvular damage in the pelvic veins, leading to a retrograde venous flow. This may occur as a result of pregnancy, family history of venous insufficiency, menopause, trauma, and deep vein thrombosis.[13,14]

Clinical Presentation

Chronic Pelvic Pain

CPP affects up to 5.7 to 26.6% of women worldwide and is responsible for 20% of gynecological referrals.[15] It is a debilitating condition that is commonly overlooked.[16] Louis Alfred Richet in 1857 was the first to coin the term "pelvic congestion syndrome" for a symptom complex consisting of chronic, dull pelvic pain, pressure, and heaviness that persists for more than 6 months without any other causes.[7] The reason behind these symptoms is thought to be due to dilated, tortuous, and congested veins in the female pelvis.[17,18] Menses, prolonged standing, and activities increasing the abdominal pressure can exacerbate the symptoms of PCS that often worsen as the day progresses.[17,18] The presence of ovarian point tenderness along with the symptom complex is highly sensitive to clinch the diagnosis of PCS.[19]

Leg Swelling/Varicosities

Varicose veins in the lower extremity related to PVI can be atypical in location due to the number of connections that exist between the pelvic veins and lower extremity veins.[7,20,21] Examples of such connections are communications between the inferior gluteal vein and the profunda femoris vein, the obturator vein and profunda femoris vein, and the internal iliac vein and great saphenous vein.[22–24] Varicosities in the lower limb usually manifest in three forms—perineal and/or gluteal varicosities, varicosities along the great or small saphenous veins, and recurrence of varicosities following treatment.

Vaginal/Labial Varicosities

Vaginal/labial varicosities are dilated and tortuous veins in the labia majora and labia minora, and are estimated to occur in 22 to 34% of women with varicose veins of the pelvis and in 18 to 22% of pregnant women.[25] Vulvar varicose veins can be associated with dyspareunia, vulvodynia, and also venous thromboembolic events, both during pregnancy and in the nonpregnant state. It is often difficult to reliably estimate the prevalence of vulvar varicosities as they remain undiagnosed in the majority of the patients due to the atypical location, personal reluctance for consultation, and in some cases due to intermittent presence of symptoms. Vulvar varicosities can develop due to anatomical connections between pelvic and external genital veins. Vulvar venous drainage into the external and internal pudendal veins deliver blood to the great saphenous vein and internal iliac veins. The veins of labia majora and minora anastomose with the uterovaginal plexus. Furthermore, the anastomosis to the pelvic veins is also via the obturator vein, superficial circumflex iliac vein, and the perforator veins from the groin, clitoris, and perineum.[26] Due to the extensive collateral venous plexuses, traditional surgical options have no significant effect on the dilated vulvar veins. The reduction of blood flow in the tributaries of the internal iliac vein will lead to the elimination of pelvic and perineal venous reflux and the disappearance of vulvar varicosity.[27]

Diagnosis

Doppler Ultrasound (Standing Position)

Duplex ultrasound (US) is the initial imaging modality for patients presenting with signs or symptoms of PVI. US is widely available and has the added benefit of no radiation exposure.US can be utilized not only for appropriate venous identification but also to visualize the venous system for thrombosis, stenosis, and/or incompetence. Transvaginal US can also be used to visualize pelvic veins which could be identified as multiple dilated hypoechoic structures around the uterus and ovaries. Color Doppler can be used to analyze the arterial and venous flow in real time. The criteria used to make the diagnosis of PVI include but not limited to tortuous and dilated pelvic veins with diameter greater than 4 mm, and low-velocity blood flow (< 3 cm/s).[17] US scanning protocols should include assessment in the supine and standing positions with the Valsalva maneuver. During upright positioning or Valsalva maneuver, Doppler waveforms in these dilated pelvic veins may reverse due to the presence of reflux.[28] The left ovarian vein can be evaluated for size and reflux with either a transabdominal or transvaginal approach. The major limitation of the US is that it is operator dependent, and patient factors like body habitus and the presence of bowel gas can lead to nonvisualization of the pelvic structures. Normal findings on the US exam cannot, therefore, be used to eliminate the diagnosis of PVI.

Computed Tomography Venography

Computed tomography (CT) venography is useful in evaluating venous anatomy, venous dilatation, and varices.[29] The role of CT venography in the workup of the pelvic venous system is limited and should be used sparingly in women of childbearing age; however, it is considered the test of choice when a structural abnormality is suspected or there is coexisting abdominal or pelvic disorder.[30] The pelvic varices appear as torturous, dilated veins in the uterine adnexa. The varicosities appear isodense to the other abdominal veins on post-contrast imaging in CT venography.[31] One of the limitations of CT venography is the demonstration of the direction of flow when compared with US, magnetic resonance imaging (MRI), and conventional venography especially in cases where the gonadal veins and the utero-ovarian arcade are filled; however, this can be mitigated with precise timing of the contrast bolus.[32]

Magnetic Resonance Venography

MR venography with a specific protocol for the evaluation of PVI has proven to be more sensitive than US.[33] Besides the added value of no radiation, MRI with multiplanar imaging

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allows for both dynamic and morphological evaluation of pelvic veins and ovarian vein reflux. The diagnostic criteria for MRI and CT proposed by Coakley et al consist of at least four ipsilateral parauterine veins of varying caliber, at least one measuring more than 4 mm in diameter or an ovarian vein diameter greater than 8 mm.[31]

On T1-weighted images, the ovarian vein and pelvic varices appear as hypointense flow voids, whereas on T2-weighted images they are hyperintense and show decreased flow velocity.[31] On contrast-enhanced T1-weighted gradient-echo MR images, dilated ovarian and pelvic varices are best visualized in the venous phase.[33] Subtracted maximum intensity projection–reconstructed images can be utilized for three-dimensional visualization of the course of dilated veins.[34,35] Retrograde filling of ovarian veins with early draining of pelvic veins into the iliac veins can be seen in the later arterial phase.

Venography with or without Intravenous Ultrasound

When the noninvasive imaging findings are inconclusive for the diagnosis of PVI or if interventional treatment is being planned, catheter venography is performed. Venographic findings that confirm the diagnosis of PVI include dilated ovarian veins with diameter $\geq$5 mm, retrograde ovarian vein reflux, uterine venous engorgement, and filling of vulvovaginal or thigh varicosities from the cross-filling of pelvic veins across the midline.[36] Intravenous ultrasound (IVUS) can be useful in detecting venous pathologies such as iliac vein compression or nutcracker syndrome which could be a cause of PVI. Beard et al have developed a venographic scoring system that includes three components: maximum diameter of the ovarian vein, time to the disappearance of contrast material, and degree of congestion.[17,37] All three components can be scored from 1 to 3 based on the severity shown in ►**Table 1**. A total score of 5 or more is found to be an objective measure of pelvic congestion, having 91% sensitivity and 89% specificity.[17]

Diagnostic Laparoscopy

Diagnostic laparoscopy is performed for patients presenting with CPP more commonly to diagnose and search for a specific diagnosis. Direct visualization can be extremely helpful to rule out other etiologies that are distinct from pelvic varices (e.g., endometriosis). There are limitations to performing diagnostic laparoscopy as it is performed in the supine position and also requires CO_2 for insufflation which further may cause compression of varices if present and therefore may result in undiagnosed disease.

Table 1 Venographic scoring system by Beard et al[17,37]

Component	Severity	Score
Ovarian vein diameter	1–4 mm (normal)	1
	5–8 mm (moderate)	2
	>8 mm (severe)	3
Time to the disappearance of contrast material after injection	0 s	1
	20 s	2
	40 s	3
Degree of congestion	Small, straight veins (normal)	1
	Tortuous veins with variable caliber (moderate)	2
	Highly tortuous veins with great variation in caliber (severe)	3

Different Causes of Varicose Veins in the Lower Extremity

Vulvoperineal Insufficiency

Vulvoperineal insufficiency is the most common variant cause of lower limb varicose veins with a reported prevalence between 4 and 8.6%.[13,38] They can be associated with PCS which occurs due to ovarian venous incompetence causing periovarian varicosities in the pelvis. Vulvoperineal varicosities extend over the buttocks and posterior thigh and are seen more commonly during pregnancy.[13] In some multiparous women, the varicosities can continue into the lower limb and connect with the great or small saphenous vein (►**Fig. 1**). These varicosities can be evaluated by using CT venography, Doppler US, or catheter-based venography. The goal of treatment in these cases is to stop the retrograde flow in the ovarian veins which can be achieved via surgical ligation, coil embolization, or sclerotherapy.[13] Once the retrograde flow is obliterated, the lower limb varicosities can be treated separately.

Intraosseous Perforating Vein Incompetence

Intraosseous perforating vein incompetence is an extremely rare cause of lower limb varicosities with a limited number of cases published in the literature.[39–41] Valvular incompetence of perforating veins draining into anterior tibia is the most commonly seen finding; however, cases with intrafibular varix are also reported in the literature.[39,40]

Imaging with CT shows intraosseous perforating vein incompetence along with varices in the soft tissues, dilated intraosseous nutrient vein, and an enlarged nutrient canal in the affected bone diaphysis.[41] Doppler US can be used to confirm the direction of venous flow and the presence of venous incompetence.[41]

Round Ligament Varicosity

Very few cases of round ligament varicosities have been described in the literature.[42,43] They are common in pregnant females and occur due to gradually progressive venous obstruction occurring due to the growth of the gravid uterus along with progesterone-induced smooth muscle relaxation.[44–46] These patients present with an inguinal mass during pregnancy and can mimic symptoms of a complicated hernia.[45] Evaluation of these varicosities is done with a combination of CT venography and Doppler US. In CT venography, an elongated venous structure running along the round ligament and connecting to the thigh varices can be seen. Doppler US helps in confirming the presence of venous incompetence.[42,44,46]

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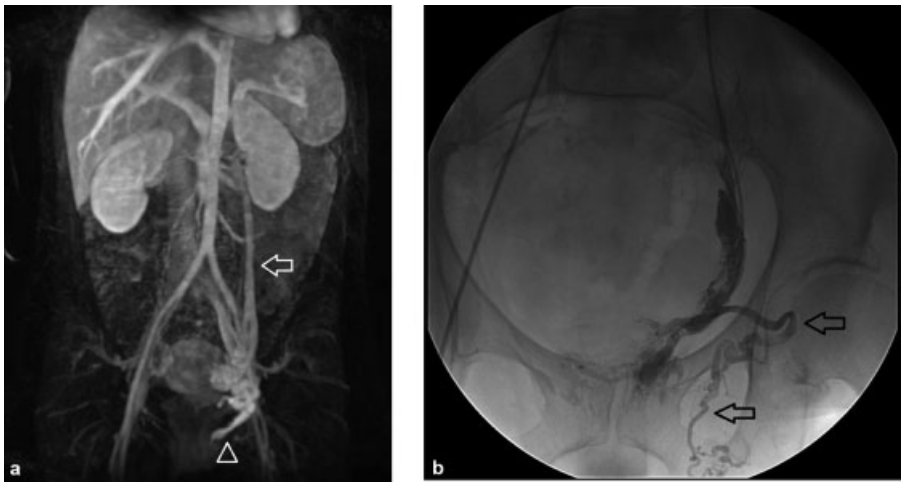


Fig. 1 Vulvoperineal varicosities due to ovarian vein reflux. (**a**) Magnetic resonance angiography/magnetic resonance venography demonstrating reflux into the left ovarian vein (arrow) and pelvic varices (arrowhead). (**b**) Catheter venography from the left ovarian vein demonstrating pelvic varices (arrows).

Persistent Sciatic Vein Incompetence

Persistent sciatic vein results from a disorder in the embryologic development of the venous system.[47] It is commonly associated with Klippel–Trenaunay syndrome (KTS) with the prevalence reported between 20 and 48% but is also seen as a primary entity.[48,49] Persistent sciatic vein incompetence presents as lower limb varicosities, and when incompetence is present below the knee, it is commonly mistaken for small saphenous vein reflux. These patients should be evaluated with a CT venography to look at the whole course and identify the connection points with varices.[50]

Klippel–Trenaunay Syndrome

KTS is a rare anomaly with a prevalence of 1 in 20,000 to 40,000 live births.[51] Patients with KTS can develop capillary malformations (port-wine stains), soft-tissue or bony hypertrophy, venous malformations, or varicose veins.[51,52] Varicosities are common in patients with KTS due to the presence of persistent embryonic veins, agenesis, hypoplasia, valvular incompetence, or aneurysms of deep veins in the superficial, deep, and perforating vein systems.[48,52] The most commonly affected leg veins are the popliteal and femoral veins.[52,53] These patients can present with pain, aching, swelling, and ulceration due to the varicosities.

The venous abnormalities in these patients can be evaluated using CT and/or catheter venography that can show abnormal superficial veins or aberrant lateral or sciatic veins along with the extent of the draining system.[53] Imaging also helps in identifying the presence or absence of varicosities extending into the abdomen or pelvis.

Portosystemic Collateral-Related Varicose Veins

There is an elevation of portal pressure in patients with portal hypertension that can lead to the opening of various portosystemic collaterals and the development of varices.[54] The coronary venous collateral varices are seen most commonly followed by esophageal varices and paraumbilical varices. The paraumbilical veins originate from the left intrahepatic portal vein and run through the falciform ligament and can be connected to the superior and inferior epigastric veins to cause the development of prominent abdominal collateral veins.[54,55] In very rare cases, lower extremity varicosities can be caused by a paraumbilical collateral pathway and only one such case has been reported in the literature.[38]

Treatment

Ovarian Vein Embolization

The earliest documented case of transcatheter embolization for incompetent ovarian vein was reported by Edwards et al in 1993 and the patient experienced extended relief of symptoms postprocedure.[56] Until recently, the main indication for ovarian vein embolization was the management of PCS due to its reported limited efficacy in improving lower limb varicosities.[57] However, in patients with refluxing pelvic veins leading to lower leg varicosities, the utilization of pelvic vein embolization has increased, but there are no adequate randomized controlled trials to support the use of this procedure.

The goal of the procedure is to target the incompetent veins which are determined with preoperative imaging MR venography or CT venography which can also be used to determine the extent of varicosities. The procedure is performed under mild analgesia and conscious sedation is not needed. First venous access is established through the jugular, arm, or femoral veins. The transjugular venous approach is most commonly used, as it is easier to gain access and offers an antegrade approach to the most frequently affected veins.

Once access is achieved, a venography of the inferior vena cava can be performed to establish the venous anatomy. The cavogram identifies any caval abnormalities, identifies the position of the renal veins, and may also show reflux into the internal iliac veins. The catheter is then advanced into the ovarian vein down to the level of the pelvic brim and a venography is done to see the reflux in pelvic veins, cross-pelvic collaterals, and pelvic varices. The embolic agent of choice is guided by operator preference and the degree of

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reflux. The commonly used embolic agents include platinum embolization coils, foam, glue, liquid sclerosants, or a combination of various embolics.[58,59] The most commonly used agent is sodium tetradecyl sulfate (STS) followed by coils.[58,59] In cases where a sclerosing agent is used, it is critical to prevent its displacement into the contralateral ovarian vein or systemic circulation. To prevent this, a balloon occlusion technique is performed by advancing the balloon into the distal ovarian vein and then advancing a microcatheter distal to the balloon. The balloon is then inflated to prevent the reflux of sclerosant around the ipsilateral ovarian veins and their crossing via the pelvic collaterals. The endpoint of sclerosant embolization is checked by increased resistance to contrast injection which is demonstrated by slow flow, stasis, or reflux of contrast. When coils or plugs are used along with sclerosant, they are deployed into the ovarian vein prior to balloon deflation. A microcatheter balloon system is then retracted proximally, and the same procedure is performed and repeated two to three times in each ovarian vein to achieve good results.

Various techniques for ovarian vein embolization have been described in the literature. Combining coil embolization with sclerosis has been demonstrated to be the most efficacious technique in multiple studies. The internal iliac veins can also be treated if they demonstrate reflux with the same technique. Both the internal iliac vein and the ovarian vein can either be treated in the same session or in multiple sessions with ovarian vein embolization first followed by pelvic vein embolization.

After the procedure, the patients can leave the hospital on the same day of the procedure. Complications associated with the procedure include coil migration into the pulmonary artery reported in 2% of cases undergoing ovarian vein embolization, perforation of the ovarian vein, flank pain, thrombophlebitis, postprocedural fevers, and puncture-site hematoma.[60–62]

Superficial Lower Extremity Varices Management

Ambulatory Phlebectomy

The concept of phlebectomy was first given by Hippocrates who hypothesized that puncturing a vein at multiple points sequentially can be used to remove the "bad blood" which feeds an ulcer. The first phlebectomy was performed by Aulus Cornelius Celsus who made large incisions along the varicose veins and then covered them with compressive bandages to allow ambulation in these patients. Ambulatory phlebectomy can be used for the management of symptomatic varicose and reticular veins; however, it is contraindicated in patients with dermatitis or cellulitis in the area of varicosities, severe peripheral edema, or those on anticoagulation or with hypercoagulable states.

Ultrasound-Guided Foam and Liquid Sclerotherapy

Injection sclerotherapy is the technique of abnormal vein destruction with the help of an agent that destroys the endothelium of the veins causing venous occlusion and fibrosis. McCausland was the first one to introduce sclerotherapy in the United States who published a case series of 10,000 patients in 1939.[63] Injection sclerotherapy is a minimally invasive procedure that can be performed in an outpatient clinic with great ease and requires very little equipment. The commonly used sclerosing agents include STS 1 to 3% (Hereford, UK) or polidocanol 0.5 to 3% (Zurich, Switzerland). Potential complications of sclerotherapy included deep vein thrombosis and thrombophlebitis.

Treatment Approach (Ovarian Vein Embolization vs. Bottom-up Approach)

The treatment approach is decided based on patients' symptoms with the most distressing symptoms driving the order of treatment. Patients with symptoms primarily related to the lower extremity or vulvar varices may first undergo management with either US-guided foam sclerotherapy or ambulatory phlebectomy in multiple sessions. However, if the patient's most distressing complaint is the pelvic symptoms, then the pelvic veins are managed first with coil embolization of ovarian veins and/or internal iliac vein sclerotherapy. Treatment of PVI in many cases can lead to an improvement in both vulvar and lower extremity varicosities. If the patient's presenting symptoms are due to underlying iliac vein compression, then stent placement may be needed with or without ovarian vein embolization.

Conclusion

Knowledge of the pathophysiology and anatomy of pelvic and superficial venous disease is important to determine appropriate treatment strategies. Preprocedural noninvasive imaging is essential for diagnosis, venous mapping, and to exclude nonvascular pathology. While superficial venous insufficiency treatment will be sufficient for the vast majority of patients presenting with lower extremity varicosities, some patients will require a more thorough investigation of their pelvic and deep venous systems.

Ethical Approval
IRB approval was not needed.

Informed Consent
Informed consent was not required for this study.

Consent for Publication
Consent for publication was not required for this study. No content of the study has been presented or published previously.

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Conflict of Interest
D.S. is a consultant for Boston Scientific and Vesper Medical.

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