

## Pudendal nerve block to treat the residual pain after embolization of pelvic varicosity in a patient of pelvic congestion syndrome

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### Abstract

Chronic pelvic pain (CPP) is a pain of at least six months' duration that occurs below the umbilicus and can cause functional disability. The clinical presentations of CPP include dull pelvic pain which is exacerbated before or during menses, dyspareunia, urinary frequency, and/or generalized lethargy. Etiologies of CPP are diverse and often needs multi-disciplinary approaches. Pelvic congestion syndrome (PCS) is a syndrome that presents with CPP and a definite anatomical findings of pelvic vein insufficiency and pelvic varices. Venography is usually used to confirm pelvic vein reflux and pelvic varices. Among the strategies to treat PCS, endovascular embolization or stent therapy, and non-pharmacologic approach, endovascular therapy has become more popular and provided favorable clinical outcome. However, there is no promise for 100% success rate without recurrence. We present the case of a 41-year-old female with CPP that first appeared when she was 36 and her left lower abdominal pain exacerbated one year ago. Gastrointestinal, urologic, obstetric and gynecological diseases were all excluded, but computed tomography angiography showed left ovarian vein insufficiency and pelvic varices. Transcatheter coil embolization of the left gonadal vein was successfully performed with no retrograde flow immediately after embolization and symptoms improved. However, the residual pelvic and perineal pain after the endovascular embolization therapy did not respond to analgesics. She received the ultrasound-guided left pudendal nerve injection 4 months later, and her pain was almost completely abolished. She was delighted with the outcome at a 6-month follow-up. This case highlights the importance of interventional pain procedure in the management of pelvic pain especially when the conventional therapeutic approaches failed.

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### Key Words:

Embolization, pelvic congestion syndrome, pudendal nerve, ultrasound, varices

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## ***I*ntroduction**

Chronic pelvic pain (CPP) refers to pain of at least six months' duration that occurs below the umbilicus and can cause functional disability. It is a common medical problem that prevales in 14-24% of women in reproductive age and in nearly 15% of women during their lifetime worldwide [1]. The potential etiologies of CPP are diverse and reflect the multiple organ systems present in the pelvis including gynecological, gastrointestinal, urologic, musculoskeletal, vascular, and body-wide disorders. The differential diagnosis of CPP includes endometriosis, pelvic inflammatory disease (PID), cancer, and pelvic varices that need mutli-disciplinary approaches. The association of CPP with pelvic varices was initially revealed by Taylor in 1949 [2], and it is now commonly referred to as pelvic congestion syndrome (PCS) with a typical age of presentation ranging from 20 to 45 years [3]. Belenky et al. identified a 9.9% prevalence of retrograde flow in the left ovarin vein (ovarian vein insufficiency) in healthy female kidney donors after preoperative abdominal aortography, and 59% in pelvic pain patients [4]. Nevertheless, it is considered to be a potential cause of CPP, and is prevalent in 10-30% of patients presenting with CPP. These findings suggest that a majority of patients with ovarian vein reflux have pelvic pain, and PCS is responsible for CPP when pelvic insufficiency results in painful pelvic varicosities [4,5].

PCS is CPP secondary to definite anatomical findings including pelvic vein insufficiency and pelvic varices [6]. The clinical presentations of PCS include dull pelvic pain which is exacerbated before or during menses, dyspareunia, urinary frequency, and/or generalized lethargy. Pain of PCS is most severe at the end of the day, exacerbated by prolonged standing, sitting and heavy activity, and is relieved by lying down. Pelvic

examination might demonstrate cervical motion and ovarian point tenderness [5]. Imaging studies with either computed tomography (CT), magnetic resonsnce imaging, transabdominal and/or transvaginal ultrasonography, or laparoscopy have all been advocated to the diagnosis of pelvic pain, but their sensitivity for pelvic varicosity is low [7]. Venography is usually necessary to confirm pelvic vein reflux when a suspicion for PCS exists, and findings include renal vein reflux into dilated ovarian vein (diameter > 5 mm), stagnation of contrast in pelvic veins, contralateral reflux across the midline of abdomen, and demonstration of vulvoperineal or thigh varices [5,8]. The combination of symptoms of CPP and positive findings of pelvic vein insufficiency and pelvic varicosity confirmed by venography come to the diagnosis of PCS.

Strategies to treat PCS have been proposed as surgical ligation of pelvic varices and ovarian vein, endovascular therapy of pelvic insufficient vein with either stent placement, embolization with coil deployment or the combined sclerosants injection, pharmacologic ovarian suppression with medroxyprogesterone acetate or goserelin (a gonadotropin releasing hormone agonist), and hysterectomy and oophorectomy [9]. However, the success rate for PCS treatment with favorable clinical improvement is around 70-90% and a recurrence rate of 12.5% [5,9]. In addition, improvement of CPP might be delayed 6 months later when pain intensity is severe at presentation [5]. Therefore, analgesics, physical therapy, neurostimulation, lifestyle change, psychological treatments, and interventional pain procedure have been suggested be provide additional pain reduction in PCS patients [5,10].

Although medications, surgery, transcatheter therapy, and rehabilitation therapy have been proposed to manage the PCS, yet no definite solution has been proposed to reduce residual pain after these approaches, especially

the pelvic vein embolization. Accordingly, this report provides an interventional pain management with ultrasound-guided pudendal nerve block to treat the residual pain in a patient with PCS after endovascular embolization therapy.

## **C**ase Report

A 41-year-old gravida 2, para 2, woman presented with a 5-year history of ambient pelvic pain that had worsened in these 6 months. Past medical history was a history of two elective caesarean sections with the last one being performed 9 years ago due to cephalopelvic disproportion and no other medical conditions. Besides, her menstrual cycle is regular with interval of 28 days. During that pregnancy, she had developed hemorrhoids which had not resolved post-delivery. The pain was a dull left lower abdominal pain which was worse with prolonged sitting, exercise and at the end of the day but was relieved after changing position. On her worst days, the pain score was 5-6 on a 0-10 point visual analog scale (VAS) for pain, and was not reduced by non-steroidal anti-inflammatory drugs including celecoxib, ibuprofen, diclofenac, and ketoprofen. Multidisciplinary survey exclude the gastrointestinal disorders, PID, gynecological disorders, and pelvic cancer. Abdominal CT scan with intravenous contrast revealed engorged left gonadal vein and PCS was therefore impressed (Fig. 1A). She was referred to the Interventional Radiology clinic for the appointment of endovascular therapy 2 months later. Intravenous digital subtraction angiography (IV-DSA) of the left renal venogram under Valsalva maneuver demonstrated retrograde flow within the dilated left gonadal vein and the appearance of pelvic varices (Fig. 1B and 1C). Under the diagnosis of PCS, transcatheter embolization with 6 Nester® Embolization coils (Cook Medical Europe LTD., Limerick, Ireland)

to occlude the left gonadal vein (8-14 × 3, 10-14 × 3). The immediate follow-up left renal venography and left internal iliac venography after coil deployment revealed no evidence of retrograde flow of left gonadal vein after embolization (Fig. 1D and 1E).

Two weeks after the endovascular embolization therapy, her moderate pelvic pain was improved but conversely she reported intermittent sharp and tingling pain at her left pelvis and perineum. Meanwhile, this pain was exacerbated especially in squat position or going to toilet. Similarly, the analgesic regimen of gabapentin (100 mg po tid), tramadol/acetaminophen (1# po tidprn), and celecoxib (200 mg po qd) was inadequate for pain relief with a VAS score 4/10 at the worst time. Therefore, she was referred to our Pain Management clinic 4 months later, for a interventional procedure consultation. Under the impression of left pudendal nerve entrapment, the ultrasound-guided techniques for pudendal nerve block at the level of ischial spine was performed with the patient in the prone position. After aseptic preparation of the patient's left buttock with betadine and alcohol, a low-frequency (2-5 MHz) curvilinear C60xp transducer connected to an ultrasound machine (Sonosite X-Porte; Fujifilm Sonosite, Bothell, Washington, USA) is placed in the transverse plane to identify the ischial spine. A 23-gauge, 7-cm needle was inserted inplane from the medial end of transducer and advanced in a medial to lateral approach until the needle tip was adjacent to the ischial spine tip. Eight mL 0.25% bupivacaine mixed with 5 mg dexamethasone was slowly injected with intermittent aspiration at the entrance of the Alcock canal and it almost eradicated her pain. Most days she only occasionally experienced very mild pelvic pain with VAS pain scores decreasing from 4 to 1 which did not require analgesic treatment. She was delighted with the outcome at 6 months follow-up.

## ***D*iscussions**

PCS is a syndrome combined with clinical presentations of CPP and anatomical findings of pelvic vein insufficiency and varices. Pelvic vein insufficiency can be the consequence of either primarily venous valve incompetence and/or secondarily to venous obstruction by the overlying artery such as May-Thurner syndrome (compression of the left common iliac vein by right common iliac artery) and Nutcracker syndrome (compression of the left renal vein between the abdominal aorta and the superior mesenteric artery). Numerous pelvic venous plexuses are relatively valveless and incompetent [11,12], and therefore, the pelvic veins are liable to dilate and even become varicose.

Surgical ligation of pelvic varices and gonadal vein, endovascular therapy, pharmacologic ovarian suppression, and hysterectomy and oophorectomy have been applied to treat pelvic pain and pelvic vein insufficiency in patients of PCS. Besides, estrogen manipulation to reduce venous distention, analgesic treatment, and interventional pain procedure are also important for symptomatic improvement. In our patient, sharp perineal pain appeared after the endovascular embolization therapy of left gonadal vein and was not effectively reduced by analgesic treatment. This pain was exacerbated especially in squat position or going to stool. Diagnosis pudendal nerve entrapment was made by clinical symptoms both with previous pelvic pain worsen with prolonged sitting and present left perineal pain. We, therefore, performed the ultrasound-guided left pudendal nerve block and successfully abolished her pain. It had been shown that pudendal nerve could be compressed by pelvic varices [13]. Consequently, pudendal nerve entrapment might be due to either pelvic plexus compression or peritoneal adhesions secondary

to previous cesarean sections in our patient and its symptoms were masked by the more prominent pelvic pain. Careful history taking and physical examination were mandatory to the successful treatment of PCS especially when mixed with peripheral nerve neuropathy. We suggest that the mechanism underlying the pudendal nerve intervention in the present case might be analgesia and/or hydrodissection release of the adhesions or compression.

## ***C*onclusion**

Treatment for patients with pelvic congestion syndrome deserves a multi-disciplinary approach not only for its complicated anatomical changes in the pelvic cavity but also for a constellation of symptoms. Peripheral nerve compression by the pelvic veins or varices could happen in patients of PCS, and its presentations should be carefully evaluated to avoid misdiagnosis.

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### ***Figure Legends***

Figure 1 (A) Abdominal computed tomographic scan with intravenous contrast revealed dilated left gonadal vein (arrow) with an estimated diameter of 1.3 cm. (B) Left renal venography demonstrated left gonadal vein insufficiency with dilatation (arrow) during Valsalva. (C) Left renal venography under Valsalvar maneuver demonstrated left gonadal vein insufficiency with dilatation (arrow) during Valsalva and later the appearance of pelvic varices (solid arrow). (D) The immediate follow-up left renal venography after coil pack (filled arrowhead) deployment revealed no reflux of left gonadal vein. (E) The immediate follow-up left internal iliac venography after coil pack deployment also revealed no reflux of left gonadal vein.

## 會陰神經介入性治療顯著改善骨盆腔鬱血症候群病人 在接受骨盆曲張靜脈栓塞後的殘餘疼痛

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慢性骨盆腔疼痛是常見的問題，持續超過 6 個月下腹部痛，可伴隨性交疼痛、頻尿或是全身倦怠，造成社會功能失調。骨盆腔鬱血症候群是慢性骨盆腔疼痛，合併有骨盆腔靜脈曲張與功能不全。治療骨盆腔鬱血症候群方法，包括用藥物抑制卵巢，外科手術切除子宮卵巢，經血管管腔內介入性治療放置栓塞線圈或是血管支架，以及非藥物治療等；但無法保證完全治癒，且不再復發。我們提出這個 41 歲女性個案，慢性骨盆腔疼痛長達六年，且骨盆腔疼痛在最近一年內更趨嚴重。排除掉消化系統、泌尿道以及婦產科問題，電腦斷層血管造影發現左側卵巢靜脈功能不全與骨盆腔靜脈曲張。經血管管腔內介入性治療，將左側卵巢靜脈成功以線圈栓塞，骨盆腔疼痛立即得到改善。但是兩周後左側會陰部刺痛與殘存的下腹部疼痛困擾她，鎮痛劑治療無效，以超音波導引進行左側會陰神經介入治療，疼痛幾近消失，六個月後她對於治療效果十分滿意。本文強調介入性止痛治療在慢性骨盆腔疼痛的重要性。

**關鍵字：** 栓塞，骨盆腔鬱血症候群，會陰神經，超音波，靜脈屈張

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