Pelvic Congestion Syndrome Presenting as Persistent Genital Arousal: A Case Report

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ABSTRACT

Introduction. Persistent sexual arousal is defined as an intrusive and unwanted genital arousal unrelated to sexual desire and typically unrelieved by one or more orgasms.
Aim. We report a patient presented with persistent sexual or genital arousal.
Methods. A 62-year-old woman presented with a 5-month history of persistent genital arousal. The symptoms were unrelated to sexual stimuli but were relieved incompletely and temporarily by orgasm.
Results. Doppler ultrasound demonstrated multiple pelvic varices. Magnetic resonance imaging (MRI) demonstrated varices in the pelvis, the vaginal wall, perineum, inguinal region, and anterior abdominal wall. Coil embolization of the dilated incompetent left ovarian vein was performed with marked reduction of symptoms.
Conclusion. We proposed that the syndrome of persistent genital or sexual arousal in women warrants a careful and systematic examination for pelvic venous incompetence by Doppler ultrasound and MRI or computerized tomography, with appropriate tilt positioning and Valsalva maneuver, before assigning it to a central neurologic or psychologic etiology. **Thorne C, and Stuckey B. Pelvic congestion syndrome presenting as persistent genital arousal: A case report. J Sex Med **;**:**–**.**

Key Words. Persistent Sexual Arousal; Persistent Genital Arousal; Pelvic Congestion Syndrome; Pelvic Varices; Ovarian Vein; Embolization

Introduction

Pelvic congestion syndrome is a condition characterized by the congestion of the pelvic veins caused by retrograde flow via incompetent ovarian veins to varices within the pelvis. The flow through the internal pudendal, obturator, and ischial veins may cause vulval congestion. Pelvic congestion syndrome typically occurs in parous, premenopausal women and is exacerbated by pregnancy, sexual intercourse, standing, and any maneuver that increases intra-abdominal pressure. The relationship of pelvic congestion syndrome to chronic pelvic pain was outlined by Beard who, in 1984, systematically demonstrated an increased prevalence of pelvic varicosities in women with pelvic pain syndrome compared with normal women and those with demonstrated pelvic pathology as a cause for their pain [1].

Persistent sexual arousal is defined as an intrusive and unwanted genital arousal, e.g., tingling, throbbing, pulsating, unrelated to sexual desire and typically unrelieved by one or more orgasms [2,3]. The characterization of persistent sexual arousal disorder, also called persistent genital arousal disorder, has focused upon the psychologic concomitants of the condition. The cause is thought to be unknown in most cases.

We report a case of a woman presented with typical symptoms of persistent sexual arousal disorder in whom ovarian vein incompetence and pelvic congestion were identified as the probable etiologic factors.

Methods

A 62-year-old Caucasian woman, gravida 4, para 3, presented to her family physician with a 5-month history of persistent genital sexual arousal. The symptoms were associated with urgency, frequency, and nocturia up to five times per night. The feeling of sexual arousal was persistent but varying in intensity. The arousal was experienced as unrelated to any subjective sense of sexual excitement or desire, and was intrusive and unwanted. In daytime, it was worsened by sitting, standing, and walking, and was partially relieved by lying down. However, she was also woken at night by feelings of sexual arousal. Orgasm, both from self-stimulation and with her husband, lessened the arousal temporarily. Although she had used self-stimulation to orgasm as a means of relieving the symptoms, she preferred not to do so regularly as the relief was temporary. The arousal was also lessened temporarily by micturition. There was an associated supra pubic and left iliac fossa discomfort.

One month prior to the onset of symptoms, she had been referred to a general surgeon for the investigation of severe anal pain. He had performed a colonoscopy and diagnosed an anal fissure. This was treated unsuccessfully initially with nitroglycerine ointment then by bupivacaine injection, with good therapeutic effect. She was also referred to a urologist who performed a cystoscopy to investigate microscopic hematuria. This was reported as showing mild trigonitis. Initially, Atrobel (Fawns and McAllen, Croyden, Vic, Australia) (hyoscine, atropine, and hyoscyamine) then carbamazepine were prescribed for bladder irritability and nocturia, with no therapeutic effect. Then, corn silk tea was prescribed as an anti-irritant. She was then referred to a practitioner who is experienced in sexual medicine.

She had been taking postmenopausal hormone replacement therapy, estradiol hemihydrate (1 mg) and norethisterone acetate (0.5 mg) (Kliovance, Novo-Nordisk, Baulkham Hills, NSW, Australia), for some months for hot flashes and osteopenia, and fluticasone inhaler for asthma. The patient was a happily married farmer's wife with good relations and communication with her husband. Her husband was supportive and caring, and attended some of the consultations with his wife. There were no identified stressors preceding the presentation of the symptoms. However, she was distressed and anxious about her symptoms to the point of being passively suicidal.

On physical examination, there were no masculinizing features, and the vulva, vagina, and clitoris were all normal to inspection, with no signs of atrophy. The vulva had normal touch and pain sensation on testing. Full blood examination and ESR, sex hormones including androgens (testosterone, free androgen index, androstenedione, DHEAS, and 17-OH progesterone), sex hormone binding globulin, and prolactin were all within the normal range as were urine microscopy and vaginal microscopy for pathogens.

Results

The pelvic ultrasound showed an anteverted uterus with an endometrial thickness of 2.5 mm. Both ovaries were seen and measured 2 cc in volume. There were multiple varices seen in the pelvis particularly in the paracervical region. Ultrasound of the left groin demonstrated multiple varicosities in the left inguinal region, some arising from the left sapheno–femoral junction, others from the vulval region.

Magnetic resonance imaging (MRI) with gadolinium revealed a dilated left ovarian vein. There were varices in the medial aspect of the thigh anteriorly near the great saphenous vein and posterior to the great saphenous vein course. Further varices were noted in the region of the sapheno–femoral junction extending to the left aspect of the perineum. Some extension was evident onto the anterior abdominal wall. These abnormalities were contiguous with extensive varices of the entire vaginal wall. Vaginal varices were contiguous with the prominent parametrial veins, more markedly on the left. A lesser degree of varices formation was noted extending from the perineum to the right inguinal region (Figure 1).

A diagnosis of ovarian vein incompetence with pelvic congestion was made. Before referral to a vascular surgeon, a second opinion was sought from a gynecologist, who described the patient's symptoms as a feeling of fullness in the pelvis, heightened sensitivity of the clitoris and vulva, clitoral swelling, and an overall feeling of sexual arousal not associated with sexual desire. He agreed that the findings on the MRI could be the cause of the patient's symptoms.

Percutaneous endovascular occlusion of the left ovarian vein was performed, using stainless steel coils with attached synthetic fibers and 3% ethoxysclerol (Figure 2). Following this, the patient noticed a marked, estimated at 70%, improvement in her symptomatology over the next 6 weeks, in

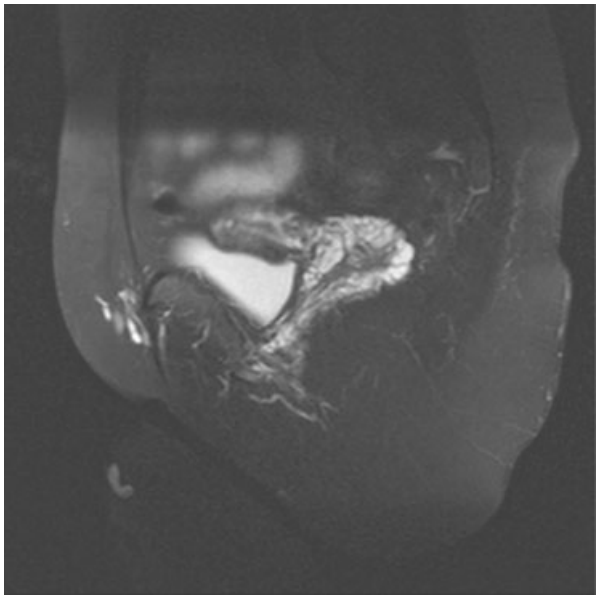

Figure 1 Magnetic resonance imaging scan with contrast enhancement shows extensive varices involving the entire vaginal wall, contiguous with the prominent parametrial veins. Varicosities are also seen in the anterior abdominal wall and in the anterior thigh.

the feeling of sexual arousal, the left iliac fossa discomfort, and bladder irritability. This improvement has continued to the time of writing. The feeling of increased sexual arousal occurred now only on exertion in the upright posture, e.g., while doing housework or while walking vigorously.

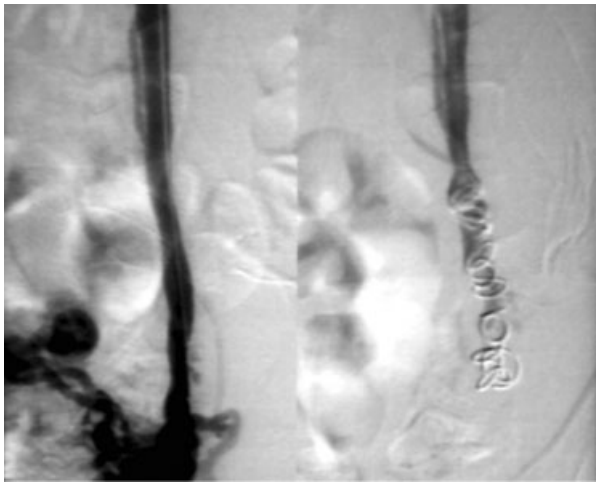

Figure 2 Venogram of the left ovarian vein shows retrograde flow and pelvic varices before embolization (left) and after embolization using stainless steel coils and 3% ethoxysclerol (right).

These were now modifiable by lying down and avoiding heavy housework.

Discussion

We reported a case of pelvic venous incompetence and extensive pelvic varices where the presenting symptoms were those of subjective persistent sexual arousal and not of pelvic pain. Imaging showed extensive pelvic varices. Given the marked appearance of the varices and the relatively short duration of the presenting symptoms, it is possible that the preceding treatment with nitroglycerine for anal fissure pain caused vascular dilatation and induced the symptoms.

Pelvic pain syndromes in women are common and have a varied etiology [4]. Many authors over many years have described the association between ovarian and pelvic varices with chronic pelvic pain [5]. It mostly arises from incompetence in the ovarian vein, mainly occurring on the left. Predisposing factors are multiparity, premenopausal status, and the anatomical drainage of the left ovarian vein to the left renal vein where it may become compressed between the aorta and the superior mesenteric artery. Retrograde flow through incompetent valves may create varices in the broad ligament, the vulva, and sometimes in the medial anterior thigh through extensive venous anastomoses. Although the usual presenting symptom is pelvic pain, there may be associated irritable bowel and bladder symptoms, as reported by our case. Symptoms are often exacerbated by prolonged standing and by sexual activity. However, persistent sexual arousal has not been reported previously as the presenting symptom of pelvic venous incompetence.

Pelvic venous incompetence and pelvic varices may be demonstrated by venography, by Doppler ultrasound, and by MRI. Filling of varices or retrograde flow may not be demonstrated if studies are done in the supine position only and Valsalva maneuver or tilting to an upright position may be necessary to reproduce the conditions under which the patient experiences the symptoms.

Venography, which involves the catheterization of the ovarian veins via a transfemoral or transuterine approach, has given way to less invasive techniques [1]. Doppler ultrasound is now the usual initial investigation to identify pelvic varices, dilated pelvic veins around the uterus and ovary, and slow or retrograde flow usually through the

ovarian veins. Computerized tomography (CT) and MRI with gadolinium enhancement may be used to demonstrate pelvic varices [6,7]. Dilated varices may be seen around the uterus and ovary, in the broad ligament, and the paravaginal venous plexus, as in our patient.

However, MRI studies have shown passive reflux of the left ovarian vein in asymptomatic women [8]. Similarly, studies using CT have found incompetent and dilated ovarian veins in 47–63% of healthy parous women with no symptoms of pelvic pain [9]. Therefore, it is possible that the attribution of causality to the vascular findings may result in inappropriate or ineffective treatment of pelvic symptoms.

The treatment of pelvic pain has, in the past, ranged from psychologic therapy to hormonal therapy. However, with the clear demonstration of venous reflux and pelvic varices, as in our case, the current treatment of choice is endovascular coil embolization of the ovarian vein. It has been reported that 50–90% of patients with pelvic congestion syndrome find that their symptoms are improved after venous embolization [10–12]. Not all patients have a complete resolution of symptoms, as in our patient, presumably because of the incomplete embolization of the tributaries and the myriad of collateral venous connections and residual venous pathways remaining untreated.

Although pelvic congestion is not uncommon, persistent sexual arousal disorder or persistent genital arousal disorder is considered to be relatively rare, or at least rarely reported. The largest series, collected from an Internet recruiting survey, has been reported by Leiblum et al. [13–15]. The characteristic of the syndrome as reported by those who responded to the survey was a feeling of genital vasocongestion that was persistent, unwelcome, and largely unrelated to sexual stimulation. However, masturbation to orgasm, often repeated, was used by many as a means of relieving the symptoms. Many respondents expressed a sense of embarrassment or shame in their symptoms. These sentiments were expressed by our patient. We propose that these feelings of embarrassment may lead to underreporting of the arousal symptoms in many patients with pelvic venous congestion.

Although the symptoms of pelvic venous incompetence and persistent genital arousal disorder are very similar, studies of pelvic vascular function in the latter condition have not been reported until now. Our patient presented with persistent genital arousal, had clearly demonstrable pelvic varices on MRI and ultrasound, and had a clear therapeutic response to embolization.

Conclusion

We proposed that the syndrome of persistent genital or sexual arousal in women warrants a careful and systematic examination for pelvic venous incompetence by Doppler ultrasound and MRI or CT, with appropriate tilt positioning and Valsalva maneuver, before assigning it to a central neurologic or psychologic etiology. We would propose that further research be conducted into pelvic venous competence in women who present with persistent sexual arousal.

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Conflict of Interest: None declared.

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