

ABSTRACT

Introduction: Persistent genital arousal disorder (PGAD) is the female perception that they are in a state of sexual arousal, without the ability of the arousal to be satisfied by orgasm.

Aims: It is the hypothesis of this study that PGAD results from a minimal degree of nerve compression of the dorsal branch of the pudendal nerve. If this hypothesis were true, then PGAD could be treated by neurolysis of the dorsal branch of he pudendal nerve.

Methods: A retrospective chart review was carried out from 2010 through 2018, of those women having neurolysis of the dorsal branch of the pudendal nerve. Patients were included in the cohort if they had a diagnosis of PGAD. All patients were assessed for demographic and clinical differences. Comparison between the pre-operative and postoperative groups was performed with descriptive statistics.

Main Outcome Measures: The changes in clitoral symptoms (arousal, numbness, pain) evaluated post-operatively.

Results: Of the 8 women included in this study, 7 were followed more than 24 weeks since surgery. Six of these women had the surgery bilaterally, and each of these had an excellent result (100%), meaning elimination of the arousal symptoms, pain, and the ability to resume normal sexual intercourse. The patient with unilateral decompression of the dorsal branch of the pudendal nerve had some improvement in arousal symptoms. This patient was the only reported case of symptom persistence.

Clinical Implications: Provides a new treatment approach for patients with PGAD.

Strength & Limitations: The main strength of the study is that this is the first article to report treatment of PGAD with neurolysis of the dorsal branch of the pudendal nerve. The main limitations of the study are the sample size and analysis of retrospectively collected data.

Conclusion: The relief of arousal symptoms supports the hypothesis that PGAD is due to a minimal degree of compression of the dorsal branch of the pudendal nerve.

INTRODUCTION

The diagnosis of the rare condition, persistent genital arousal disorder (PGAD) requires female patients to meet the following five criteria: 1) genitals are persistently aroused, 2) arousal remains following orgasm or requires multiple orgasms to diminish, 3) arousal is unrelated to desire, 4) arousal is triggered by both sexual and non-sexual stimuli, and 5) symptoms are intrusive and unwelcomed.¹⁻³ Unwelcomed or unwanted arousal has been observed as the most common detrimental symptom in 91.3% of women surveyed in the largest study identifying PGAD symptom characteristics. The majority of these women are unable to derive emotional satisfaction from sex.

The hypothesis being tested in this study is that the underlying mechanism causing PGAD is a minimal degree of chronic compression of the dorsal branch of the pudendal nerve. This is the nerve supplying sensory innervation to the clitoris and is often referred as the dorsal nerve to the clitoris.¹⁶ In 1980, it was reported that a minimal degree of chronic nerve compression was interpreted as a hypersensitive response to a vibratory stimulus when applied to the ulnar nerve in patients with cubital tunnel syndrome (Table 2).¹⁷ With more advanced degrees of nerve compression, there was decreased perception of the vibratory stimulus.¹⁷ In 2016, using a rat model of chronic nerve compression, it was also observed that the lesser degree of nerve compression resulted in a lowering of the sensory (pain withdrawal) response, indicating a lower threshold (hypersensitivity) with a minimal degree of nerve compression.¹⁰

The present clinical study evaluates the response of women with PGAD to decompression (neurolysis) of the dorsal branch of the pudendal nerve.

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MATERIAL AND METHODS

Patient selection

Inclusion criteria were female patients 1) referred for PGAD of more than 52 weeks duration, who 2) had been treated already by their Gynecologists for all known medical causes of this disorder, who 3) have had already pelvic floor therapy, who 4) have had a trial of neuropathic pain and anti-anxiety medication treatment, and who 5) had stopped any physical activity, such as cycling, that may be known to cause compression of the pudendal nerve.

Surgical technique

The patient is placed into lithotomy position, the perineal hairs are clipped and a betadine scrub and prep are done. Loupe magnification at 3.5X is utilized. A bipolar coagulator is used at the lowest possible voltage, especially when dissecting adjacent to the dorsal branch of the pudendal nerve at the inferior pubic ramus to avoid electrical injury to the nerve. The local anesthetic used is 1% Xylocaine injected into the incision site, which is located superior to the ischial tuberosity, and at an angle to the inferior pubic ramus, and lateral to the labia majora (Figure 1A).

The incision is opened into the ischiorectal fossa and maintained open with a Weitlander retractor with blunt "teeth." Dissecting posteriorly and inferiorly, the perineal branches are identified, and preserved. In ten percent of patients the dorsal branch will exit through the canal of Alcock, ¹⁸ and so this variant must be identified. If it is present, then the scarring around the exit must be released, and this would complete the procedure. This has not proven to be the anatomic finding in any of the patients operated on in this series.

After confirming that there is no dorsal pudendal branch exiting the canal of Alcock, the Weitlander retractor is placed into the most superior aspect of the incision. In this location, the ischiocavernosus muscle is identified and its origin from the inferior pubic ramus is released using first the bipolar coagulation and then a sharp scissors. Care must be taken not to enter the corpora cavernosa to prevent venous bleeding. As the dissection approaches the juncture of the inferior pubic ramus with the symphysis pubis, a softer region is palpated, and spreading transversely with the scissors reveals a yellowish, almost fatty appearing 2 mm diameter dorsal branch of the pudendal nerve. Care must be taken not to injure electrically this little nerve. The nerve will be in slightly tight fascia as it travels more superficially to the base of the clitoris. The nerve will still likely be entrapped by scar more proximally. A small right angle clamp or dissector can be placed between the nerve and remaining intact fibers of the inferior pubic ramus canal to delineate this tight region prior to cauterizing it and then sharply releasing it under direct vision while protecting the nerve. (Figure 1B)

The site of compression can often be observed, being narrowed, and often inflamed compared to the regions proximal and distal to the compressed nerve (Figure 2,3,and 4).¹⁹

After checking for hemostasis, the wound is closed with interrupted, intradermal 4-0 monocryl, and the skin with interrupted and continuous 5-0 nylon sutures. The dressing is Xeroform, gauze, and small Tegaderm.

Figure 1



Ambulation and showering is allowed. The bandage is removed on postoperative day #3 and then betadine is applied twice a day. Sutures are removed between the 12th and 14th day after surgery.

Rehabilitation consists of water walking in a heated pool, 3 to 4 times per week for 15 minutes each time. This can be continued if desired for 3 more weeks, increasing to 30 minutes and including swimming.

Primary outcomes measured were pre- and postoperative 1) persistent arousal symptoms, 2) pain measured by visual analogue scale, and 3) numbress, with changes defined as complete relief (CR), partial relief (PR), or no relief of symptoms. Secondary outcomes measured were the change in ability to have preoperative and postoperative intercourse. Other secondary measures included, mean time to improvement of symptoms following surgery, mean length of time for patient follow-up, postoperative complications, and symptom recurrence.

Demographics Table 3 lists the demographics for all 8 women included in this study. Variables of interest include: patient age; gender; body mass index (BMI); race and ethnicity; if the patient was a current smoker, consumed alcohol or recreational drugs; marital status; number of prior gynecological surgeries for an unrelated cause; total number of pregnancies; total number of children; family history of PGAD or related symptoms; mean number of comorbidities per patient; patient comorbidities; patient occupation; the cause of initial mechanism of injury; date of first presentation to peripheral nerve specialist, mean length of time of PGAD symptoms; mean number of preoperative medications to treat PGAD symptoms at first visit.



MATERIAL AND METHODS

Figure 2



Variables and outcomes analyzed

Figure 3

•Eight women with a mean age of 51 years (standard deviation: 10 years) were included (Table 4). Seven of these women had surgery bilaterally. At a mean time of 60 weeks following surgery, 7 of the 8 women (88%) had complete relief of arousal and 1 (12%) had partial relief following unilateral decompression of the dorsal branch of the pudendal nerve. Of the 7 women that had pain, 6 (86%) had complete relief after bilateral decompression, and 1 (14%) had partial relief following unilateral decompression of the dorsal branch of the pudendal nerve. No patients reported pre-operative numbress for post-operative comparison. Five of the 6 (83%) patients unable to perform intercourse without pain were able to perform sexual intercourse without pain after surgery. The mean time to improvement of the first symptom following surgery was 11 weeks (standard deviation: 11 weeks). No major surgical complications were observed.

Figure 1



This is the first report of neurolysis of the dorsal branch of the pudendal nerve in women with PGAD. The relief of arousal symptoms supports the hypothesis that PGAD is due to a minimal degree of compression of the dorsal branch of the pudendal nerve. From the results of this study, it appears that a unilateral decompression may not be sufficient to achieve an excellent result for patients with PGAD.

Ther 2018;44:111-126. improvement. J Sex Med 2009;6:1479-1486. 2016;50:321-330 14. ICD10Data.com. 2018;2018. 23. Leiblum SR, Chivers ML. Normal and persistent genital arousal in women: new perspectives. J Sex Marital Ther 2007;33:357-373.

RESULTS

CONCLUSION

REFERENCES

1. Goldmeier D, Leiblum SR. Persistent genital arousal in women -- a new syndrome entity. Int J STD AIDS 2006;17:215-216 2. Facelle TM, Sadeghi-Nejad H, Goldmeier D. Persistent genital arousal disorder: characterization, etiology, and management. J Sex Med 2013;10:439-450. 3. Leiblum SR, Nathan SG. Persistent sexual arousal syndrome: a newly discovered pattern of female sexuality. J Sex Marital Ther 2001;27:365-380. 4. Jackowich R, Pink L, Gordon A, Poirier E, Pukall CF. Symptom Characteristics and Medical History of an Online Sample of Women Who Experience Symptoms of Persistent Genital Arousal. J Sex Marital 5. Bedell S, Goldstein AT, Burrows L. A periclitoral mass as a cause of persistent genital arousal disorder. J Sex Med 2014;11:136-139 6. Filler AG. Diagnosis and treatment of pudendal nerve entrapment syndrome subtypes: imaging, injections, and minimal access surgery. Neurosurg Focus 2009;26:E9. 7. Gaines N, Odom BD, Killinger KA, Peters KM. Pudendal Neuromodulation as a Treatment for Persistent Genital Arousal Disorder-A Case Series. Female Pelvic Med Reconstr Surg 2018;24:e1-e5. 8. Korda JB, Pfaus JG, Goldstein I. Persistent genital arousal disorder: a case report in a woman with lifelong PGAD where serendipitous administration of varenicline tartrate resulted in symptomatic 9. Kruger THC. Can pharmacotherapy help persistent genital arousal disorder? Expert Opin Pharmacother 2018:1-5. 10. Pettersson LM, Danielsen N, Dahlin LB. Altered behavioural responses and functional recovery in rats following sciatic nerve compression and early vs late decompression. J Plast Surg Hand Surg 11. Rosenbaum TY. Physical therapy treatment of persistent genital arousal disorder during pregnancy: a case report. J Sex Med 2010;7:1306-1310. 12. McMullen R, Agarwal S. Persistent Genital Arousal Disorder-Case Report of Symptomatic Relief of Symptoms With Transcranial Magnetic Stimulation. J ECT 2016;32:e9-e10. 13. Studd J. A comparison of 19th century and current attitudes to female sexuality. Gynecol Endocrinol 2007;23:673-681. 15. Markos AR, Dinsmore W. Persistent genital arousal and restless genitalia: sexual dysfunction or subtype of vulvodynia? Int J STD AIDS 2013;24:852-858 16. Pauls RN. Anatomy of the clitoris and the female sexual response. Clin Anat 2015;28:376-384. 17. Dellon AL. Clinical use of vibratory stimuli to evaluate peripheral nerve injury and compression neuropathy. Plast Reconstr Surg 1980;65:466-476. 18. Colebunders B, Matthew MK, Broer N, Persing JA, Dellon AL. Benjamin Alcock and the pudendal canal. J Reconstr Microsurg 2011;27:349-354 19. Furtmuller GJ, McKenna CA, Ebmer J, Dellon AL. Pudendal nerve 3-dimensional illustration gives insight into surgical approaches. Ann Plast Surg 2014;73:670-678. 20. Hrynko M, Kotas R, Pokryszko-Dragan A, Nowakowska-Kotas M, Podemski R. Persistent genital arousal disorder - a case report. Psychiatr Pol 2017;51:117-124. 21. Balaya V, Aubin A, Rogez JM, Douard R, Delmas V. The dorsal nerve of the clitoris: surgical applications. Morphologie 2014;98:8-17. 22. Robert R, Labat JJ, Riant T, Louppe JM, Hamel O. The pudendal nerve: clinical and therapeutic morphogenesis, anatomy, and physiopathology. Neurochirurgie 2009;55:463-469.

24. Groneman C. Nymphomania: A History. New York, NY: W.W. Norton & Company, Inc; 2000. 25. van der Walt S, Oettle AC, Patel HR. Surgical anatomy of the pudendal nerve and its branches in South Africans. Int J Impot Res 2015;27:128-132. 26. Pink L, Rancourt V, Gordon A. Persistent genital arousal in women with pelvic and genital pain. J Obstet Gynaecol Can 2014;36:324-330.