

Persistent Genital Arousal Syndrome; A Case Report

Persistan Genital Uyarılma Sendromu; Bir Olgu Sunumu

Ezgi Selcuk-Ozmen^{1,a,*}, Asli Enzel-Koc^{2,b}

¹Trabzon Fatih State Hospital, Psychiatry Clinic, Trabzon, Türkiye.

²Cumhuriyet University, Faculty of Medicine, Psychiatry Clinic, Sivas, Türkiye.

*Corresponding author e-mail: selcukezgi@gmail.com

^a<https://orcid.org/0000-0002-2057-4947>

^b<https://orcid.org/0000-0002-9853-745X>

ABSTRACT

The purpose of this case report is to present a case involving the etiology, diagnosis, and treatment of Persistent Genital Arousal Disorder (PGAD). The described patient meets the criteria for PGAD. In this case, organic lesions in the nervous and urogenital systems were excluded, and a psychogenic background of the syndrome was assumed. Due to the patient's refusal of pharmacological treatment, psychoeducation regarding PGAD and sexuality was provided within the framework of cognitive-behavioral therapy, resulting in a moderate effect.

Keywords: Cognitive behavioral therapy, Persistent genital arousal disorder, Sexual dysfunction

ÖZET

Bu olgu sunumunun amacı, Kalıcı Genital Uyarılma Bozukluğu'nun (PGAD) etiyolojisi, tanısı ve tedavisini içeren bir vakayı sunmaktır. Tanımlanan hasta PGAD kriterlerini karşılamaktadır. Bu vakada, sinir ve ürogenital sistemlerdeki organik lezyonlar dışlanmış ve sendromun psikojenik bir temele dayandığı varsayılmıştır. Farmakolojik tedaviyi reddetmesi nedeniyle, hastaya bilişsel davranışçı terapi çerçevesinde PGAD ve cinsellik konusunda psikoeğitim verilmiş ve orta düzeyde bir etki elde edilmiştir.

Anahtar Kelimeler: Bilişsel davranışçı terapi, Cinsel işlev bozukluğu, Kalıcı genital uyarılma bozukluğu

INTRODUCTION

Persistent Genital Arousal Disorder (PGAD) is a diagnosis that has gained increased awareness in recent years and is gradually being introduced into the literature. It has not yet been included in the ICD-10 or DSM-5 classifications. Since it was first defined by Leiblum and Nathan in 2001, numerous case reports describing individual cases or small groups of patients with this disorder have been published.¹ In 2019, the International Society for the Study of Women's Sexual Health (ISSWSH) published the first expert consensus on the diagnosis and treatment of PGAD.² These criteria include:

1. Unwanted or intrusive genital arousal sensations that persist for at least 3 months or longer, which may be disturbing,
2. It may also include other types of genito-pelvic dysesthesia, such as buzzing, tingling, burning, throbbing, itching, and pain,
3. Additionally, frequent occurrences in the clitoris and other genito-pelvic areas such as mons pubis, vulva, vestibule, vagina, urethra, perineal area, bladder, and/or rectum,
4. It may involve being on the brink of orgasm, experiencing uncontrollable orgasms, and/or an excessive number of orgasms,
5. Not associated with accompanying sexual desire, thoughts, or fantasies.

Patients with PGAD commonly use terms like itching, tightening, pain, or swelling to describe their symptoms. These sensations can last for hours or throughout the day, causing extreme discomfort for the patient. Due to

feelings of shame and embarrassment, patients tend to conceal their symptoms. Another challenging factor for patients is the lack of experience among many physicians regarding these symptoms and the absence of a clear consensus on treatment.

The pathophysiology and etiology of PGAD are not clearly understood. Central neurological changes (e.g., post-injury, specific brain lesion/anomaly), peripheral neurological changes (e.g., pelvic nerve hypersensitivity or compression), vascular changes (e.g., pelvic obstruction), mechanical pressure on genital structures, medication-related changes (such as the use or discontinuation of antidepressants and other mood stabilizers), psychological changes (stress), onset of menopause, physical inactivity, and overactive bladder are implicated in the etiology.¹

The frequency of the disorder has not been determined to date. Until recently, this disorder was only identified in women; however, a few cases of PGAD have also been reported in men.³ Symptoms frequently occur in women aged 25-51 and men aged 38-74.⁴

In more than 60% of PGAD cases, an overactive bladder syndrome has been found, strongly associated with Restless Leg Syndrome (RLS).¹ The relationship between PGAD and RLS is uncertain. RLS is believed to arise from iron deficiency in the central nervous system, dysfunction of the nigrostriatal system, and imbalances in dopaminergic and glutamatergic neurotransmission involving opioids and hypocretin.⁵ The neuropathogenesis of RGS proposed by Waldinger is presented in Figure 1.⁶ In this case report, a female patient diagnosed with Persistent Genital Arousal Syndrome is being discussed in line with the literature.

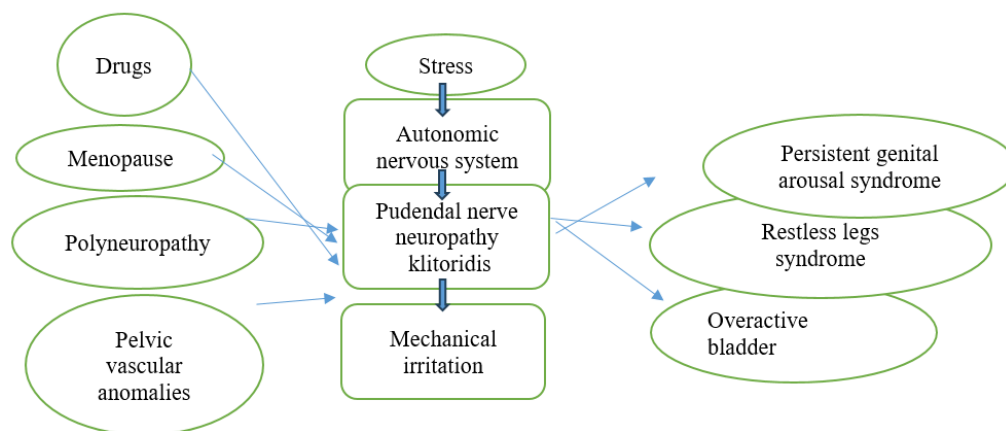


Figure 1. Diagram of the hypothetical neuropathogenesis of restless genital syndrome

CASE REPORT

A 24-year-old single woman applied to the hospital. She complained of experiencing spontaneous orgasms during daily activities lasting less than 20 minutes, occurring 4-5 times a day, without feeling sexual desire for the past 2 years. The patient mentioned feeling pain in her right groin, followed by a pulsating sensation in her vagina, along with vaginal discharge. She stated that due to the need to shower after each orgasm experience during the day, she had become unable to leave home, experiencing significant social and academic difficulties. There were no organic disorders in the patient's medical history, and she did not have a history of chronic illness, continuous medication use, alcohol, or substance abuse.

Due to her symptoms, she first consulted a gynecologist. During the urogynecological examination, it was observed that her external genital structure was in line with sex norms, her hormone levels were normal, and there were no abnormalities in the thyroid function test or biochemistry values. Her neurological examination was normal. No pathology was detected in the computerized brain tomography and Electroencephalography (EEG).

In the mental status examination, her overall appearance and physical development were in line with her socioeconomic status. It was determined that not feeling sexual desire and still being able to have an orgasm caused psychological tension and dissatisfaction.

The psychological evaluation, based on the Minnesota Multiphasic Personality Inventory (MMPI), indicated that her responses were defensive, with no clinical pathology detected.

During the period of sexual development, the patient remembered that she had not received any information about menstrual bleeding from her elders or peers before, and did not react when it first occurred. The patient stated that she had never been in a flirtatious situation, engaged in sexual activity, or masturbation. She recalled an incident at the age of 9 when she played a sexually themed game with her 19-year-old cousin's son, during which he exposed his genitalia to her. When she attempted to touch him, she felt extreme anxiety and ran away upon hearing her sister calling them from another room. The patient mentioned missing school for a month after the incident due to daily abdominal pain, and she had never shared this incident with anyone.

A followup interview was carried out with the patient about her illness. Showing limited knowledge about

sexuality, she received 2 sessions of psychoeducation. Paroxetine 10 mg (often preferred for treating anxiety and depression, regulating serotonin levels, managing sexual desire, and controlling involuntary arousal with sexual side effects) was prescribed to manage her anxiety symptoms, such as fear, restlessness, irritability, palpitations, sweating, muscle tension, and abdominal discomfort due to genital stimulation at unwanted times. A follow-up appointment was scheduled for 15 days later.

During followup interviews, the patient expressed that she did not want to use the recommended medical treatment and confirmed that she did not use it. Following psychoeducation, the patient reported a decrease in the frequency of pain in her right groin and expressed relief, feeling that there was no reason to blame herself for her complaint. The patient was advised to seek further help if symptoms increased or if she experienced additional complaints. Written informed consent was obtained from the patient who participated in this case.

RESULTS

Persistent Genital Arousal Disorder (PGAD) is a highly distressing and disgraceful condition for patients. Since its first report in 2001, clinical treatment strategies for PGAD have significantly expanded, providing a rational basis for managing this condition in many patients.⁷ However, it should be emphasized that the care process for PGAD/PGD is somewhat limited to expert opinion due to a lack of awareness about the condition and its impact, insufficient research, the absence of large-scale studies on effective treatments, and inadequate research support. The numerous and varied etiologies also hinder the development of a single treatment strategy, necessitating a personalized, biopsychosocial approach. Increasing awareness of this condition, combined with expanding clinical experience and efforts to improve patient outcomes, may lead to affected individuals achieving a better quality of life.

Benzodiazepines such as clonazepam and oxazepam are often prescribed to alleviate PGAD symptoms. The medication's mechanism of action is likely based on the 'gating' phenomenon of the Central Nervous System.⁶ Tramadol, a medication that activates μ -opioid receptors and has a limited effect on serotonin and noradrenaline reuptake, has also been documented as beneficial for some patients.¹ The tricyclic antidepressant amitriptyline has proven effective in treating patients with depressive or compulsive disorders. While researchers have explored the potential

use of dopamine receptor agonists (such as pramipexole) as a treatment for Restless Leg Syndrome (RLS) and Restless Genital Syndrome (RGS), the results have been inconsistent. The effectiveness of Botulinum toxin injections in treating pudendal neuropathy has also been demonstrated.⁸

Family therapy and couples therapy may benefit patients experiencing long-term stress due to family conflicts.⁹

In our case, the patient's psychological symptoms of stress related to preparing for a central exam, along with relief from non-pharmacological methods after receiving psychoeducation, suggest that the complaints could be psychologically driven. Some PGAD cases have shown that psychotherapy, particularly cognitive-behavioral therapy used in the treatment of genital pain syndrome, may be effective.

CONCLUSION

As there is currently no standard treatment algorithm for PGAD, a thorough psychiatric and physical medical history, along with comprehensive examinations and accompanying diagnoses, is essential in guiding treatment selection. In our case, psychotherapy was effective in reducing tension and symptoms, supporting the psychosomatic nature of the disorder. However, the etiology and treatment options for PGAD remain under-researched. Detailed assessment for PGAD symptoms is crucial, and structured research is necessary to establish effective long-term treatment approaches for this condition.

Authorship contribution statement

Concept and design: ESO and AEK.

Acquisition of data: ESO and AEK.

Analysis and interpretation of data: ESD and AEK.

Drafting of the manuscript: ESO.

Critical revision of the manuscript for important intellectual content: ESO and AEK.

Declaration of competing interest

None of the authors have potential conflicts of interest to be disclosed.

Ethical approval

No ethical approval was required for this study. Informed consent was obtained from the patient who participated in this case.

Availability of data and materials

All data generated or analyzed during this study are included in this published case.

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