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Persistent genital arousal disorder – a case report

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Summary

The persistent genital arousal disorder (PGAD) may coexist with restless legs syndrome and overactive bladder syndrome and share some similarities with these conditions. Thus, the new term: restless genital syndrome (RGS) is proposed. The purpose of this paper is to present a case of PGAD, including the description of the etiology, the diagnostics and the treatment of the disorder. The described patient meets the criteria for PGAD. Organic lesions within nervous and urogenital system were excluded and the psychogenic background of the syndrome was assumed in this case. The patient was diagnosed with personality disorder with a predominance of dependent personality traits and emotional lability. After the failure of pharmacological treatment, systemic psychotherapy with cognitive-behavioral elements was initiated with moderate effect.

Key words: persistent genital arousal disorder, restless genital syndrome, restless legs syndrome

Introduction

The persistent genital arousal disorder (PGAD) is a condition that includes an intrusive and undesired genital arousal without sexual stimulation. The disorder was first described by S. Leiblum and S. Nathan in 2001 [1]. To date, several case reports have been published, describing individual cases or small groups of patients diagnosed with this disorder [2–5]. The diagnostic criteria for PGAD were developed in 2010 and include:

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1) spontaneous genital arousal that persists for extended periods of time (hours, days, months);
2) persistent arousal that does not resolve with an orgasm;
3) arousal that is not accompanied by a sexual desire;
4) arousal that is unwanted;
5) the symptoms cause distress [6].

To date, the incidence of the disorder has not been determined. Until recently, the disorder was described only in women. However, a few cases of PGAD in men have been published as well [7–9]. The symptoms occurred in 25 – to 51-year-old women and 38 – to 74–year-old men [7, 8,10–13]. In the majority of the female cases, the symptoms commenced in the menopausal and peri-menopausal period. The majority of the patients remained in long-term relationships (60–70%) [14, 15].

The etiopathogenesis of the syndrome is unclear and appears to be complex. Psychological factors, organic causes and certain drugs and substances are thought to trigger the syndrome.

Psychological factors include sex-related emotional problems. 60% of female patients with PGAD also suffer from depressive disorders, 40% have concurrent anxiety disorders; approximately 10% experienced sexual abuse in childhood and approximately 10% have symptoms of the burnout syndrome [12]. PGAD may also be associated with pelvic vascular malformations (a high incidence of pelvic varicose-veins), a history of a stellate ganglion block, mechanical damage or irritation of the pudendal nerve or small-fibre polyneuropathy [5, 8, 15]. PGAD was also described as an adverse effect of the use or discontinuation of selective serotonin reuptake inhibitor antidepressants (SSRI) and an increased dietary intake of soy [2, 16]. 40% of the patients with PGAD had a history of alcohol abuse; the symptoms were also observed after discontinuation of cannabinoids and nicotine [10–12, 14, 15, 17].

PGAD was found to coexist with the overactive bladder syndrome (OAB) in over 60% of the reported cases and was strongly associated with the restless legs syndrome (RLS) [5]. Similarly to RLS, resting (sitting position) aggravated PGAD symptoms in approx. 70% of female patients, while movement, especially walking alleviated complaints in approx. 30% of the patients [5]. In some cases the symptoms worsened in evening and during the night hours [18]. Hence, the disorder was coined the restless genital syndrome (RGS) by some authors, which may suggest a common pathogenesis of these disorders [5, 10, 12–15]. Both PGAD and OAB were therefore thought to arise due to sensory neuropathy of the pudendal nerve and the dorsal nerve of the clitoris [15]. The association between PGAD and RLS remains unclear, since RLS is thought to occur as a result of iron deficiency in the central nervous system, dysfunction of the nigrostrial system and imbalances in the dopaminergic and glutaminergic neuro-transmission that involve opioids and hypocretin [19–23]. The neuropathogenesis of RGS proposed by Waldinger is presented in Figure 1 [15].

Attempts to treat PGAD included pharmacological therapy, psychotherapy and application of invasive procedures.

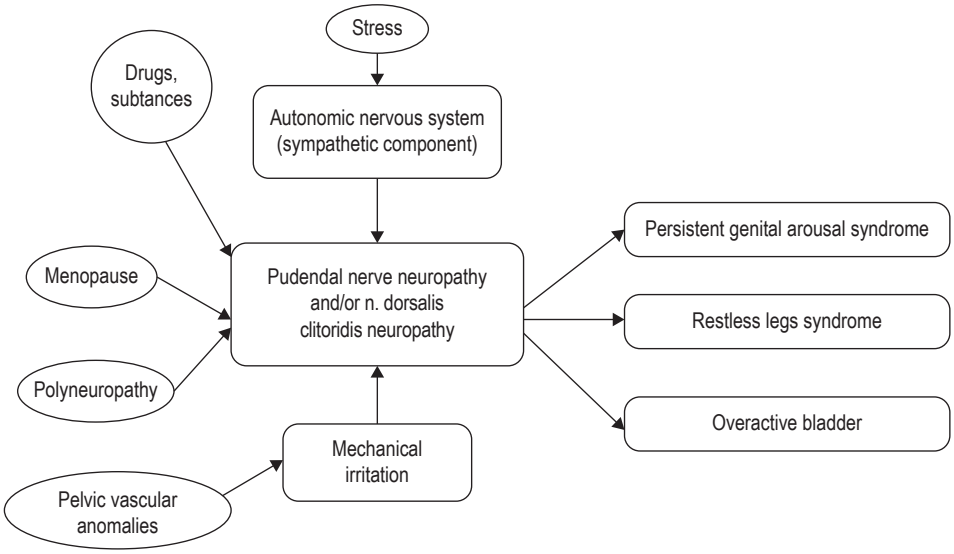

Figure 1. **Diagram of the hypothetical neuropathogenesis of restless genital syndrome** according to Waldinger et al., 2009 [15] – in own modification.

Benzodiazepines such as clonazepam and oxazepam are most commonly used to mitigate PGAD symptoms. Their mechanism of action is most likely based on the CNS "gating" phenomenon [15]. Tramadol, which is an agonist of μ-opioid receptors and a weak selective serotonin and noradrenaline reuptake inhibitor, was reported to be effective in some patients [5]. Tricyclic antidepressants, such as amitriptyline, were successfully used in patients with depressive or compulsive disorders [24, 25]. Based on the analogy between RLS and RGS, attempts were also made to treat the disorder using dopamine receptor agonists (pramipexol), without consistent effect [5]. Diagnose of neuropathy or polyneuropathy led to treatment with antiepileptic drugs, such as carbamazepine, oxcarbazepine, pregabalin and gabapentin [18]. Antidopaminergic risperidone was used in a single case [26]. Hormone replacement therapy was used in women in the postmenopausal period with different therapeutic effects [5].

In some PGAD cases, psychotherapy, including cognitive-behavioral therapy which has been used in the therapy of genital pain syndrome, may be effective. Family therapy and couple therapy may be beneficial for certain patients (e.g., subject to long-term stress caused by family conflicts) [24, 27].

Botulinum toxin injections were successfully used in patients with pudendal neuropathy [28]. Men with a coexistent neuropathy were treated with transcutaneous electrical nerve stimulation (TENS), with different therapeutic effects, and pregabalin which was effective in one case [7, 8]. Some case reports described an improvement in PGAD symptoms in patients following the embolization of pelvic varices and electroconvulsive therapy in patients with bipolar disorder [29, 30].

Due to low incidence of this disorder, there are no clinical trials to evaluate the efficacy and safety of methods of treatment listed above.

Case report

A 40-year-old, employed woman who had completed secondary education sought psychological help due to ongoing symptoms of itchiness, irritability and burning sensation of the genitals and nonsexual genital arousal that recurred for 21 years. The symptoms did not resolve with an orgasm during intercourse or masturbation. The severity of the symptoms was variable but no factors were found to evoke or affect them (not related to time of day, body position etc.). The patient first noticed the symptoms after initiating sexual relationship with her partner, to whom she has remained married for 20 years now.

The woman reported regular menstrual cycles. She used hormonal contraception for a short period of time, which she discontinued due to poor tolerability. She did not notice any correlation between an exacerbation of the symptoms and her menstrual cycle.

The symptoms were persistent and caused increasing discomfort. Occasionally, they caused sleep disorders, which decreased her concentration and led to poor performance at work. Severity of symptoms sometimes made her avoid sexual intercourse with her husband. Nevertheless, she admitted to benefit from her condition in a way, because her complaints were addressed with compassion and care by her husband and son.

The feeling of incomplete bladder emptying during voiding has reoccurred for two years. Apart from this, the patient did not suffer from any other urological or gynecological disorders. Other complaints included tension-type headaches that occurred for many years, and were occasionally treated with non-steroidal anti-inflammatory drugs. In the year prior to the consultation, the patient had a one-month history of alcohol abuse. She did not report any other substance abuse. The family history revealed that the patient's father had an alcohol dependence syndrome and that he imposed his religious beliefs on the whole family.

During examination the contact between the physician and the patient was constructive; the patient had good verbal fluency, was auto – and allo – psychologically oriented and did not display any productive symptoms. Her mood was variable, with features of depression, drive was normal. The patient attempted to precisely answer all the questions, although she was tense and anxious. Despite the occasional sleep disorders reported by the patient, which have been mentioned above, at the time of the examination the patients reported that she slept well. She had normal appetite. On neurological examination no symptoms and signs of damage to central or peripheral nervous system were found, especially features of (poly)neuropathy, radiculopathy or myelopathy, which could explain the described symptoms. Exception for bladder problems, no other signs or symptoms suggesting autonomic dysfunction were reported (e.g., vasomotor abnormalities, abnormal pupillary response, orthostatic changes in blood pressure, intestinal dysfunction). In order to exclude the subclinical lesions in peripheral and autonomic nervous system, electrophysiological diagnostics, including

electroneurography, somatosensory evoked potentials, and an examination of sympathetic skin response and cardiac variability were planned. However, the patient did not agree to perform any of the above-mentioned tests.

The results of psychological tests (Structured Clinical Interview for DSM-IV Axis II Personality Disorders – SCID-II; Raven's Progressive Matrices – standard version; Benton Visual Retention Test; Graham–Kendall Memory-for-Designs Test) revealed that the patient had average intellectual abilities with no signs of organic changes in the central nervous system; furthermore, the tests indicated features of a dependent, emotionally unstable personality with a tendency toward anxiety and depression [31–34].

The computed tomography (CT) did not show any lesions within the brain. The results of gynecological examination, as well as CT and ultrasound examination of the abdomen and pelvis were normal. Laboratory tests revealed slightly increased anti-thyroid peroxidase antibody (anti-TPO) titters that reached 295 IU/ml with normal TSH, fT3 and fT4 values. The urine tests revealed a recurrent urinary tract infection caused by E.coli.

Gynecologist and neurologist conducted the initial management of the patient. Due to lack of organic lesions she has been under the care of the Outpatient Mental Clinic for 5 years. The patient was preliminarily diagnosed with a neurotic disorder. Venlafaxine was administered at a daily dose of 150 mg for several months, but caused itching and unpleasant genital sensations. A combined therapy including sertraline at a dose of 200 mg/day and alprazolam at 1 mg/day was administered in the following year, without significant improvement. A subsequent therapy (escitalopram at 10 mg/day and doxepin at 100 mg/day) was discontinued by the patient due to a lack of effect. The patient was unwilling to receive further pharmacological treatment, but agreed to psychotherapy. The family systemic therapy and elements from cognitive-behavioral therapy (cognitive, experience-based and relaxation techniques), were used. Establishing a longer, safer patient-psychologist relationship was of great importance for the treatment process. Periodically, when the patient experienced a re-aggravation of symptoms, there was a need to increase the intensity of meetings to two per week. Conversation with a therapist, and above all the possibility of being heard and the sense of acceptance caused partial relief. This technique resulted in gradual reduction of the perceived tension, and thus alleviation of most of the symptoms.

Discussion

Based on the nature and circumstances of the symptoms and the subjective patient's attitude, the present case meets all PGAD diagnostic criteria [6].

Similarly to other case reports, our patient was diagnosed with a personality disorder with a tendency toward anxiety and depression, episode of alcohol abuse, and a family history suggesting emotional problems in childhood and adolescence. Our patient reported a relatively early (as compared to other reports) onset of symptoms (at approximately 19 years of age), which persisted for several years [12, 13]. At the same time, she remained in a long-term relationship. Co-existing micturatory ailments may suggest overactive bladder syndrome, however, taking into consideration the

results of the laboratory tests and lack of other symptoms typical for OAB (urinary frequency, urinary incontinence, urgency) a urinary tract infection cannot be ruled out. The patient did not present with symptoms of RLS. Apart from tension-type headaches (which may have been associated with emotional distress) she did not suffer from any co-existing conditions. The diagnostic examinations did not reveal any abnormalities in the genitourinary and nervous system, including autonomic component. Hormonal abnormalities were also excluded. Increased serum levels of anti-TPO with normal thyroid hormone levels call for further investigations into possible autoimmune thyroid disorders. However, owing to the duration of the symptoms, an association between PGAD and thyroid dysfunction is very unlikely.

Pharmacotherapy did not alleviate the perceived tension and other symptoms reported by the patient, while psychotherapy aided in the reduction of tension and other symptoms, which also indicates a psychosomatic nature of the disorder. Nevertheless, due to emotional lability, pharmacological treatment stabilizing mood could support psychotherapy. Benzodiazepines lowered the level of anxiety. Discontinuation of alprazolam use was possible after introduction of antidepressant therapy.

The described case meets the diagnostic criteria of PGAD, which seems to be associated only with psychological factors. The continuation of psychotherapy and possible future pharmacotherapy may alleviate the symptoms and improve functioning of our patient.

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