

# Persistent genital arousal disorder. The case of a 35-year-old patient

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## Abstract

**Introduction:** The Persistent Genital Arousal Disorder is a relatively recently described and rarely diagnosed sexual dysfunction. In the study of its causes, the role of vascular and neurologic factors is raised. In this article an attempt has been made to conduct a comprehensive case conceptualization of a 35-year-old female patient with PGAD, taking into account the opportunity of interaction of biological and psychological factors.

**Material and methods:** An analysis has been made on the basis of diagnostic material including neuro-imaging, gynecological and psychological examinations as well as data from medical history, observation and a resume written by the patient.

**Results:** A number of overlapping factors that can constitute the etiology of a disorder as well as exacerbate its the symptoms have been identified in the patient. They are: generalized atrophy in the CNS, mixed features of personality disorders of cluster B with hysterical and conversion mechanism of exacerbations and abundant venous plexus in the labia majora.

**Conclusions:** The Persistent Genital Arousal Disorder and its symptomatic exacerbations can be complex as far as their causes are concerned and may require a broad diagnostic and therapeutic perspective. Interdisciplinary approach seems to have some potential in explaining individual mechanisms of the disorder.

**Key words:** PGAD, personality, treatment

Polish Sexology 2016; 14 (1): 46–51

## Introduction

Persistent genital arousal disorder (PGAD) was described in 2001. The authors presented 5 cases of women with very similar symptoms of unwanted sexual arousal. They called this syndrome „Persistent sexual syndrome“ PSAS [1]. In subsequent years this syndrome was described as PGAD. What is more, the exact criteria for the diagnosis [2,3] were provided. The diagnostic criteria include:

1. Involuntary feeling of stimulation of the clitoris which continues for a long time (hours, days, months).
2. Physical feeling of arousal does not disappear after one or more orgasms (obtained also after the physical stimulation of the clitoris).
3. Genital stimulation is not associated with the subjective feeling of sexual desire.

4. Persistent feelings of sexual arousal is felt as intrusive and unwanted.

5. Persistent genital arousal causes anxiety.

The year 2009 saw the publication of works suggesting frequent co-occurrence of PGAD symptoms with restless legs syndrome (RLS) and symptoms of the overactive bladder (OBS). The study on 18 Dutch women with symptoms of PGAD showed symptoms of restless legs also in the past. Waldinger called this syndrome “restless genital syndrome” REGS [4, 5]. Pudendal nerve neuropathy, and particularly excessive stimulation of sensory fibers of the dorsal nerve of the clitoris, was assessed as the main cause of the combination [6].

REGS criteria include:

1. Undesirable and unpleasant feeling of sexual arousal without feeling sexual desire or without sexual fantasies.
2. Unwanted or unpleasant sexual orgasms or sexual fantasies.
3. Symptoms of Restless Legs Syndrome.

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Received: 10.03.2015

Accepted: 12.12.2015

4. Symptoms of the overactive bladder.
5. The intensification of unwanted and unpleasant feelings of sexual tension while sitting.
6. Unsolicited hyperesthesia to sensory examination in the genital area.
7. Unwanted and unpleasant sexual experiences as a result of the examination below the pubic bone.

PGAD is therefore a new diagnosis, not in the ICD-10 or DSM-V [7]. The currently used diagnosis may be called REGS/PGAD [8].

Thus, PGAD is a new diagnosis, which is not present in ICD-10 or DSM-IV R. There are no epidemiological data, no animal models, no precise clinical trials. Also, the treatment is based on individual cases. There is also no research with a double-blind trial.

The etiology of the syndrome is based on several hypotheses.

They can be divided to:

1. The above-mentioned problem of the pudendal nerve neuropathy.
2. Vascular factors — PGAD is to be secondary to pelvic venous congestion syndrome and the problems of varicose veins in genitals [9].
3. Central neuropathy — symptoms of PGAD are secondary to the symptoms of CNS damage or to surgeries on arteries and jugular veins and interventions in the lumbar region. Symptoms are similar in their mechanism as the symptoms of epilepsy or Gilles de la Tourette syndrome [8].
4. Hormonal agents — particularly increase of the levels of dopamine and serotonin [10].
5. Pharmacological factors — use of Trazodone, rapid withdrawal [11].
6. Psychogenic factors — increased levels of anxiety, depressive symptoms [12].
7. Other — using soy (phytoestrogens) [13].

It could be stated that all the factors mentioned describe the potential for exacerbation of symptoms in individual cases. Currently, PGAD have also been diagnosed in men [14]. Therefore, the treatment is based on case descriptions. Methods of treatment include pharmacology and physiotherapeutic methods. Psychotherapy, primarily the cognitive-behavioral one, is also important [15]. Use of benzodiazepines (clonazepam and oxazepam), tramadol, locally bupivacaine [6], neuroleptics (especially the ones increasing prolactin level) were described. Rapid improvement on olanzapine was described [16]. A similar reaction of improvement was observed after 2 weeks of taking varenicline in 49-year-old female patient with a long-term, virtually lifelong, PGAD syndrome [17]. Use of tricyclics, duloxetine and pregabalin [18].

It is postulated to withdraw trazodone, soy, venlafaxine as well as to use and withdraw SSRIs. [19] There are

descriptions of cases in which PGAD was treated with electroconvulsive therapy in the case of simultaneous occurrence of bipolar disorder [20]. Physiotherapeutic methods are based on transcutaneous stimulation of the pudendal nerve, both in women [21] and in men (in 1 case without success) [22]. Surgeries in the case of vascular problems [23]. The article presents long-term PGAD syndrome in a female patient with borderline personality who was for a long time treated for depression and addiction to masturbation. Personality traits of the patient, strong mechanisms of denial and lack of diagnosis of PGAD resulted in the lack of therapeutic success.

## Case description

Patient, aged 35, divorced, having a 6-year-old son cared for by her husband, a graduate of a hotel industry school, currently on a rehabilitation allowance, came to the clinic because of the increase in the severity of the symptoms of sexual tension, which, as she stated, prevented her normal functioning in society and at the workplace. She was born in due time, from second pregnancy and second birth, without complications. Normal psychomotor development. When the patient was 2.5 years old her father went abroad. He did not visit her and her mother in Poland and the patient did not have any contact with him until she was 6 years old. He appeared at the time as, according to the patient's feelings, "Mr. Perfect Stranger". In her childhood, from about 5 years of age she suffered from "restless legs syndrome". "Strange legs" bothered her until 17 years of age, but she has never undergone any pertinent treatment. When she was 7 years old her younger brother was born, which made her feel rejected, of little importance and very upset that the mother devotes her time to him. At 13 years of age the patient had her first period. She was terrified and did not know what was happening to her, because at her home the issues related to sexuality were never discussed. Since then, she menstruates relatively regularly and without pain. She started her sexual life at the age of 15 and despite that, as she stated, it did not give her any pleasure and she feared pregnancy, "she felt that her soul's deep desire is to love and be loved" so she decided that she should sacrifice herself by having sex with her partner. The boy whom she was meeting was an alcoholic. He had problems with the law, he "stole and lied", beat her, but she felt she was very much in love with him and she was planning to spend her life with him. This relationship had a very stormy course. The patient reported that she had continued emotional swings and temper tantrums during the relationship. Due to the difficulties in coping with emotions she experimented

with alcohol and marijuana. As a teenager, she was rebellious, confrontational, she was making rows at home and she often beat her younger brother. Because of her problems with learning she had to change her school. After graduating from high school, she took a job as a baby sitter while continuing her education in the hotel industry school. During the time of her education she entered fleeting relationships several times, but they did not give her satisfaction. At the age of 20 she entered a masochistic relationship with a "philosopher who taught her his way of looking at the world". When this relationship ended she was in a deep depression. She had two suicide attempts at that time, the first time she wanted to drown herself in the Vistula River and then she attempted to hang herself (no one knew about these attempts except for one close female friend). At that period masturbation, as she states, helped her to fall asleep and quieten her thoughts. Therefore, she masturbated virtually every evening. At the same time, the patient noticed that the symptoms of sexual tension without experiencing feelings of desire began to appear every two weeks and lasted for several days. At the age of 23 masturbation in order to relieve the tension began to take her about 2 hours a day. Due to the growing problem associated with the permanent feeling of strong and long-lasting tension in the clitoris, the patient started sex therapy. She was prescribed Androcur, after which she had as many as "up to 100 orgasms a day and she could not withstand it." After the interrupted Androcur treatment, masturbation took her already about 4 hours a day. Because of this, the patient started to attend meetings for people addicted to pornography and masturbation. During this time, she met her future husband. He gave her a sense of security, but she did not love him and being with him did not give her pleasure. She felt the pain during intercours with her husband while masturbation brought her relief associated with a feeling of sexual tension. To survive she cut off from her emotions and she did not care what will happen to her, she felt as if "closed". At the age of 27 the patient began psychiatric treatment. She received antidepressants, valproate medicines, pernazinum and benzodiazepines with little effect ("physical pressure no longer appeared daily but three times a week"). In 2007, due to the increase in the severity of symptoms associated with genital arousal the patient underwent psychiatric hospitalization with the following diagnosis: other personality disorders F 60.8. During her stay in the ward the patient masturbated between ten and twenty times a day in order to get rid of the symptoms of peripheral genital arousal, which she considered a pathological and reprehensible behavior. She was discharged before the end of treatment due to lack of therapeutic

effects. Another psychiatric hospitalization took place in 2011. During the whole stay in the ward the patient was undergoing sex therapy. After psychological examination made with the use of the MMPI-2 the patient was diagnosed with emotionally unstable personality. The gynecological examination dated 2011 showed no deviations from the norm. During the whole stay the patient received pharmacotherapy including Amitriptyline (dose: 75 mg/d.), individual therapy, relaxation training sessions and biofeedback. The patient was discharged with a diagnosis of borderline personality and the Persistent Genital Arousal Disorder, in a stable mental state, without signs characteristic to the PGAD. According to the patient, the improvement remained over a period of approximately one and a half months after the end of hospitalization and it was the longest period without symptoms from the time of the onset of persistent genital arousal. After completion of hospitalization the patient decided to file a lawsuit for divorce because, as she stated, her "husband became unbearable". She experienced extreme emotions alternately. She claimed that she did not love her husband, but that he was a good man and that she decided on this relationship because she wanted to have the feeling of being in a normal family. After the divorce she was in several relationships that did not bring her satisfaction. She describes her partners as persons who for various reasons did not engage mentally in the relationship or invalidated it. The patient remains conflicted with her mother and currently has a partner who is addicted to gambling and who she is planning to move in with. The patient came to the clinic with the expectation of "fixing herself" and said that if the stay would not help her she would decide on surgical intervention (removal of the labia and clitoris). She reported a sense of lack of emotions, a sense of a weak sense of who she is, difficulties in entering close relationships with people. It is clear that initially she had difficulty in speaking about intimate topics, describing her difficulties as "somatization symptoms". She described her symptoms as "purely physical". She reported the need for compulsive stimulation of the clitoris without a subjective feeling of sexual desire, which persists despite one or more orgasms and which is experienced by her as onerous and unwanted. For this reason the patient, as she reports, is experiencing severe anxiety, fear, and so negative emotions "that she made herself feel only 20% of them". She reported that she attended therapy, which, as she claims, does not help her whatsoever. Because of the "somatization disorders" she currently receives a rehabilitation allowance. She speaks about her difficulties as "my illness", "my ordeal". In the past the patient had episodes of bulimia, which were intensifying when she was experiencing the feelings of an-

**Table 1.** Results of selected psychological examinations

Method	Numerical results
MoCA	27 pts./30 pts.
RAVLT	AI: 6, AII: 10, AIII: 12, AIV: 12, AV: 13, BI: 6, AVI: 13, AVII:13
ROCFT	copy: 36 pts., 100 percentyl; typ 2, print: 17 pts., 20 percentyl, typ: 1.
FAB	FAB: 18 pts./18pts.
TMT A i B	TMT: A: 47 sec., 10–25 percentyl, B: 56 sec., 50–75 percentyl
Stroop	A: 44 sec., B: 62 sec., C:95 sec.
MMPI-2	F = 59, Fb = 52, Fp = 70, FBS = 55, L = 70, K = 67, S = 62 // Hs = 52, D = 68, Hy = 70, Pd = 69, Pa = 54, Pt = 48, Sc = 52, Ma = 49
Verbal Fluency Test	F: 12, A: 14, S: 14, animals: 15, sharp objects:12
BSL-23	BSL-23: 12 pts.; 0 pts.

Abbreviations in the text

ger. In periods when she felt a large level of tension and stress there also occurred short-term (lasting several hours) psychotic decompensations. The patient demanded pharmacological treatment in the ward. She confirmed the symptoms associated with impaired interpersonal functioning: poor ability to recognize the feelings and needs of others, interpersonal oversensitivity (tendency to feel neglected and insulted by others), the relations which she established with others were marked by mistrust, she presented a tendency to idealization and devaluation with alternating engagement in and withdrawal from the relationship. During hospitalization the patient focused on her lack of sense of security, on her helplessness and uncertainty. Typically, the patient was in “gloomy mood”. She confirmed a sense of shame and lower self-esteem associated with the necessity to discharge tension through masturbation. In the past she regressed into deep depression or infantile dependence and she treated masturbation as, *inter alia*, a form of coping with emotions (mainly anger) and to punish herself. Severe anxiety was observed in the patient. She also reported nervousness, tension, fear, often in response to interpersonal stress, worrying about the negative effects of recent unpleasant experiences and future possible negative experiences, fear of losing control. The patient reported fear of rejection, anxiety associated with a sense of excessive dependence and loss of autonomy. In connection with the reported by the patient symptoms of the Persistent Genital Arousal Disorder, an extended neuroimaging and neuropsychological diagnosis have been performed with respect to the patient. In the MRI of the head made in the SE sequence in the T1 weighted image as well as in the TSE sequence in the T2 and TIRM images and DWI resolve and GRE 3D T2 the following was revealed: the correct structure of the brain and cerebellum without focal lesions in all applied sequences.

The psychological included the following diagnostic methods: 1) the Montreal Assessment Cognitive,

MoCA, equivalent 7.2 version, 2) The Rey Auditory Verbal Learning Test (RAVLT) with 15 words, 3) The Rey-Osterrieth Complex Figure Test (ROCFT), 4) Frontal Assessment Battery, 5) Test of joining TMT A and TMT B points, 6) Stroop word-color interference test, 7) Minnesota Multiphasic Personality Inventory MMPI-2, 8) Sachs and Sidney Sentence Completion Test, 9) Verbal Fluency Test, 10) BSL-23 list of Borderline Symptoms and 11) Female Sexuality Questionnaire — expanded Kroměříž scale. Below are the numerical results of individual studies.

The results obtained in test examinations indicated a generally proper cognitive functioning of the patient. Verbal and language learning processes developed at the average level in the patient. The patient may have a little difficulty in structuring and organization of memory material, which could potentially disrupt the efficiency of extracting it from memory. Inhibitory control processes and the processes of controlling concurrent operations do not exhibit pathological features.

The results of the MMPI-2 control scales indicate the patient's tendency to deny the difficulties and moderate tendency to report unusual pathological symptoms.

The results of the clinical scales of the MMPI-2 indicate that the patient has a tendency to depressive mood, which may worsen when she does not experience interest from other people. The patient may show many symptoms reflecting unconscious emotional states. Probably the patient may have limited insight into her emotional functioning and the nature of her problems, she may look for her sense of identity in other people and seek their interest but avoid going into deeper relationships. Probably she is able to feel the pleasure associated with closeness and intimacy to a little extent. The probable mechanism of increase in the reported symptoms is similar to the classically understood conversion. In view of the limited psychologizing abilities of the patient, her ability to benefit from the therapy is questionable. Instead of conscious anxiety

ety the patient feels tension, fatigue, weakness and may have difficulty with starting and taking up activities. The patient may be interested in status, power and rivalry, but is afraid to be in a situation of direct competition herself. Likewise, she may feel the need for greater accountability and change, but avoids the associated tension. She may experience excessive suffering from even the slightest criticism. In the emotional sphere she probably experiences a sense of "emotional blocking", she denies the occurrence of unacceptable impulses and fears her own spontaneity. The sense of discomfort may worsen especially in the company of the opposite sex. Therefore, she is capable of feeling pleasure from intimate relations with the opposite sex to a negligible extent. The patient has complex personality difficulties reflected in the expression of unconscious conflicts and deficits in the form of symptoms, with simultaneous reduced psychologizing capacity. The symptomatic mechanism can be defined within the framework of the classic understanding of the concepts of hysteria-conversion.

In connection with the fact that literary sources raise the issue of the significance of vascular lesions in the pelvic and mechanical pressure on genital structures in the etiology of the Persistent Genital Arousal Disorder, an additional MR test of the pelvic was made and gynecological consultation was suggested. The pelvic MR test (10.02.2014) made in the SE sequence in the T1 weighted images and in the TSE sequence in the T2 and TIRM images the following results were obtained: anteflexed uterus, homogeneous myometrium, cervix without changes, the outlines of the pelvic cavity are equal, endometrial is uniform and thin (3 mm), OC, right ovary: 31 × 20 mm with follicles up to 5mm, left ovary 24 × 18 mm with follicles up to 4 mm, the correct quantity in the pouch of Douglas, abundant venous plexuses in the labia majora.

Gynaecological consultation. The patient menstruates regularly, without pain. The patient takes Microgynon from 2008.. Gynecological operations (-). Vulva in the study without deviations. PV: white secretion in the vagina, abundant, vaginal activity is formed, the canal is closed. Corpus uteri is of normal size, hard, movable, painless. Appendages on both sides without perceptible lesions. Peritoneal symptoms are negative. The performed laboratory examinations showed results within the normal range, with the exception of elevated levels of prolactin (TSH — 2.107  $\mu$ IU/ml, prolactin — 40.09 ng/ml, FSH — 11 mIU/ml, LH — 10 mIU/ml, testosterone — 40 ng/dl). Treatment of patients included psycho-education and pharmacotherapy. During the entire stay the patient remained in individual therapy according to the cognitive-behavioral protocol. At the

same time pharmacotherapy with olanzapine 5 mg/d. and fluoxetine at a dose increasing to 60 mg/d. was implemented. The symptoms disappeared in the period in which the patient chose to express anger with her partner, which resulted in another separation initiated by him. Shortly before discharge from the hospital, the symptoms appeared again. This was concurrent with the patient coming back to her partner. At the same time the mechanism of denial activated in the patient — she more often claimed that the symptoms have no relation with psychological states and occur in her in an unpredictable way. However, the overall level of symptoms and their frequency were noticeably reduced. Also the overall level of the patient's psychosocial functioning increased: she was less focused on symptoms, she decided to move out from her mother's place and found prospects in starting her own business.

## Discussion

In the present case, symptoms of chronic sexual arousal syndrome have been observed in the patient for many years. Because of these symptoms the patient has been diagnosed with addiction to masturbation, which resulted in taking a therapy for addicts in Sexaholics Anonymous groups. Indeed, some PGAD symptoms may suggest symptoms of addiction, i.e. loss of control over sexual behaviors and regulation of tension and emotions through masturbation. Also, few studies have shown that some patients may have a problem with addiction [12]. However, PGAD symptoms were more primitive. The suffering was accompanied by traits of an emotionally unstable personality (borderline type), which worsened the symptoms especially during increased mental tension.

Repeated attempts of outpatient treatment gave no results. Today it is not known what is a primary diagnosis of the female patient, but certainly personality disorders intensified the symptoms. In the literature we did not find examples of the influence of borderline personality traits on the symptoms of PGAD. It thus seems that this diagnosis, which is relatively new and almost unknown to the doctors and therapists, should be taken into account especially after the diagnosis of diagnostic criteria of PGAD. The patient has the history of symptoms of restless legs syndrome and diagnostic tests confirmed the symptoms of abundant venous plexi in the labia major. The use of olanzapine and fluoxetine and work on the relations with the partner (expression of anger to him) resulted in the recession of symptoms of sexual arousal. The similar response of the patient took place in a hospital in 2011 (amitriptyline and sex therapy were used). The appearance of symptoms was

observed at the moment of discharging from hospital and, consequently, at the moment of quick contact with her partner, which, when accompanied by a strong denial mechanism (the patient denied the relationship between emotions and intensification of sexual symptoms) gives a picture of the classical understanding of the concepts of hysteria and conversion.

## Conclusions

Based on the comprehensive assessment of medical history, sexological and psychosocial assessment, performed observation and executed diagnostics, the patient has been diagnosed with the persistent genital arousal disorder with a complex etiology and borderline personality with narcissistic and histrionic (cluster B) personality traits.

The presented case shows the need to take account of personality traits, psychological mechanisms and physical factors in the diagnosis of PGAD syndrome. Since we do not have complete information on the etiology and the treatment methods of PGAD, it is difficult to assess what is the basic mechanism of the emerged symptoms which put patients at risk of long-term suffering. With the coexisting problems of addiction to masturbation it is advisable to consider that the patient may be suffering from PGAD syndrome and to establish a concrete diagnosis. This is associated with a different method of treatment. It is also worth to propagate more intensively the new and less-known syndromes related to the sexual area, with a full presentation of these dysfunctions as part of a larger whole, and not an isolated sexual symptom. Still, the treatment and diagnosis of PGAD syndrome will depend on the intuition of a doctor and the trial and error method [23].

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