UDC 616.89-008.481-008.6:618.17]-036-079.4-08
DOI: 10.14739/2310-1237.2023.1.270216

Persistent genital arousal disorder: clinical, differential diagnostic and care aspects (clinical report)

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A – research concept and design; B – collection and/or assembly of data; C – data analysis and interpretation; D – writing the article; E – critical revision of the article; F – final approval of the article

Key words: PGAD, diagnosis, treatment, care, clinical report.

Pathologia 2023; 20 (1), 94-98

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Persistent genital arousal disorder (PGAD) is a condition of persistent genital arousal, in the absence of a psychological component and organic pathology from the vascular, endocrine and nervous system.

The aim of the study is to examine the symptoms, clinical picture, developmental dynamics, risk factors, and treatment for PGAD.

Materials and methods. The clinical case of observation of PGAD in a 20-year-old female patient suffering from recurrent depressive disorder is presented. Methods included in clinical, psychopathological and psychodiagnostic examination have been with using the following techniques: Patient Health Questionnaire-9 (PHQ-9), Generalized Anxiety Disorder-7 (GAD-7), Visual Analogue Scales of pain intensity (VAS).

Results. As a result of collection of life and disease history, analysis of examination findings by related specialists and additional research methods of patient with PGAD the next diagnosis has been made according to the International Classification of Diseases 10th Revision: Recurrent depressive disorder, current episode of moderate severity with somatic symptoms (F33.11). She has been treated with a number of drugs from the Selective serotonin reuptake inhibitors group and mood stabilizers, the combination of sertraline and lamotrigine appeared the optimal treatment option. The results of the treatment have been satisfactory, the symptoms of PGAD have stopped bothering the patient, the emotional state has improved significantly.

Conclusions. The case of the PGAD is presented. Clinical picture of the case turned out to be typical in comparison with the results of other researchers. The peculiarity is the possible association of symptoms with withdrawal of escitalopram. The case confirms the data upon the high suicidal risk in this disorder. The patient's treatment requires intervention of different medical specialists to rule out organic pathology and using of multidisciplinary approach. Sertraline and lamotrigine appeared effective in the treatment of PGAD.

Ключові слова: розлад постійного генітального збудження, діагностика, лікування, організація допомоги, клінічний випадок.

Патологія. 2023. Т. 20, № 1(57). С. 94-98

Розлад постійного генітального збудження: клініка, диференційна діагностика та особливості допомоги (клінічний випадок)

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Розлад постійного генітального збудження (англ. Persistent genital arousal disorder, PGAD) – стан безперервного генітального збудження без психологічного компонента збудження й органічної патології з боку судинної, ендокринної та нервової систем.

Мета роботи – вивчення симптомів, клінічної картини, динаміки розвитку, факторів ризику та методів лікування розладу постійного генітального збудження.

Матеріали та методи. Наведено клінічний випадок власного спостереження розладу постійного генітального збудження в пацієнтки віком 20 років, у котрої діагностовано на рекурентний депресивний розлад. Дослідження передбачало застосування клінічних і психопатологічних методів, здійснення психодіагностичного обстеження з використанням Patient Health Questionnaire-9 (PHQ-9), Generalized Anxiety Disorder-7 (GAD-7), візуальної аналогової шкали інтенсивності болю (ВАШ).

Результати. У результаті збору анамнезу життя та захворювання, аналізу висновків обстеження суміжними спеціалістами та даних додаткових методів дослідження встановили діагноз за Міжнародною класифікацією хвороб 10 перегляду: рекурентний депресивний розлад, поточний епізод помірної тяжкості з соматичними симптомами (F33.11). Пацієнтка отримувала лікування низкою препаратів із групи селективних інгібіторів зворотного захоплення серотоніну та стабілізаторами настрою; оптимальною схемою лікування виявилась комбінація сертраліну та ламотриджину. Результати лікування задовільні, симптоми розладу постійного генітального збудження перестали турбувати пацієнтку, її емоційний стан значно покращився.

Висновки. Наведено випадок доволі рідкісного в медицині розладу постійного генітального збудження, клінічна картина якого виявилася типовою порівняно з результатами досліджень інших авторів. Особливість наведеного клінічного випадку розладу постійного генітального збудження полягає в можливій асоціації симптомів зі скасуванням есциталопраму. Цей випадок підтверджує відомості про високий суїцідальний ризик при розладі постійного генітального збудження. Курація пацієнтки потребувала втручання фахівців із різних галузей медицини (неврологія, гінекологія та психіатрія) для виключення органічної патології та забезпечення мультидисциплінарного підходу. Під час лікування цього випадку розладу постійного генітального збудження ефективними виявилися такі препарати, як сертралін і ламотриджин.

Persistent genital arousal disorder (PGAD) is a condition of persistent genital arousal, in the absence of a psychological component of arousal and organic pathology from the vascular, endocrine and nervous systems. For the first time the term of PGAD has been mentioned since 2006, but until today this problem has not been deeply researched and studied, so the data were presented by case studies and expert opinions [1,2]. According to some reports, the worldwide prevalence of this disorder is 0.6–3.0 % among women, but this disorder remains largely unrecognized by medical practitioners. Given the above, nowadays, practicing clinicians encountering patients with this disorder are forced to be guided by scant information and individual recommendations, which affect the quality of care [3,4]. Most of the patients do not have access to the care they need at all. Considering the severity of the symptoms of this disorder, the level of the patients' maladjustment, the accompanying symptoms from the mental sphere, and the high risk of suicide, this subject requires coverage and study, followed by recommendations for medical practitioners.

Aim

To examine the symptoms, clinical picture, developmental dynamics, risk factors, and treatments for PGAD.

Materials and methods

The clinical case of observation of persistent genital arousal disorder in a 20-year-old female patient suffering from recurrent depressive disorder is presented.

To determine mental status of the patient we conducted clinical and psychopathological study, to assess level of symptoms of depression – Patient Health Questionnaire-9 (PHQ-9) [5], anxiety – Generalized Anxiety Disorder-7 (GAD-7) [6] and pain – Visual Analogue Scales of pain intensity (VAS) [7].

In accordance with the Declaration of Helsinki, written informed consent to undergo a psychiatric examination was received from the participant.

Clinical case

Patient complaints. A 20-year-old female patient asked for help complaining of unpleasant sensations in the external genital area, mainly in the clitoral area, describing them as feelings of arousal, tension and itching, which are not pleasant and not associated with sexual arousal. They occur spontaneously during the day with the maximum intensity of these symptoms observed when the patient takes sitting or horizontal position. This state of arousal was accompanied by secretion of lubrication, pulsating and undulating sensations, blood filling of the genitals, which sometimes led to spontaneous orgasms that did not bring relief, but instead increased the arousal. Touches in the external genital area were not pleasant and resembled an "electric shock". The presence of these symptoms in the patient was accompanied by an increase in the level of anxiety and feelings of guilt: "This is the worst thing that's ever happened to me in my life", "it's like a 24/7 self-rape".

At maximum symptom severity, there was pain that spread to the external genitalia and perineum, which could only be relieved by taking a warm bath.

There was a direct correlation of these symptoms with the severity of anxiety.

These symptoms completely maladjusted the patient. They were accompanied by sleeping disorders due to painful and unpleasant sensations, decreased mood, irritability, anxiety, suicidal thoughts, and a desire for surgical treatment of this problem due to unbearable feelings.

Anamnesis. The patient was born in a complete family, there is no information about mental disorders among her relatives, she grew up and developed without deviations, the family is complete and religious. At school she studied well. Currently, she is a high school student.

At the age of 15, according to the patient, there was an attempted rape, she reported: "the boyfriend on the date began to getting handsy, tried to touch her, which frightened her and she ran away", after which she no longer engaged in a relationship and was wary of men. Around the age of 16, she first sought help from a psychiatrist regarding an anxiety disorder and was prescribed standard therapy with selective serotonin reuptake inhibitors (SSRI) antidepressant and a tranquilizer. Further, the patient periodically asked for help in connection with an aggravation of anxiety and depressive symptoms, standard therapy with antidepressants of SSRI class (fluoxetine, paroxetine) and tranquilizers was prescribed. The patient also sought help from a psychotherapist during this period of time.

For the first time, symptoms in the genital area occurred a year ago (autumn 2021) after the withdrawal of the antidepressants of the SSRI class (escitalopram). According to the patient, these symptoms began with discomfort in the urethra when urinating and were similar to those of cystitis. Further, discomfort in the clitoral area and sensation of arousal on the verge of orgasm appeared, but the patient did not inform the attending physician and psychotherapist; after 2 weeks, due to the presence of anxious-depressive symptoms, escitalopram at a dose of 10 mg was prescribed, after that the genital symptoms disappeared. However, the therapy of targeted affective symptoms was not effective enough, then a change of the treatment regimen was carried out: the patient was taking sertraline 75 mg + lamotrigine 25 mg + aripiprazole 5 mg. Given regimen allowed to keep the satisfactory affective state, however, some depressive and anxious symptoms persisted.

Subsequently, the patient began to visit a psychotherapist, who canceled this regimen and prescribed vortioxetine (March 2022), after which the patient's emotional condition worsened somewhat. Further, the psychotherapist canceled the vortioxetine (May 2022), the patient's emotional condition was stable. However, the patient did not see the effect of the visit to the therapist, so she discontinued visiting him.

This deterioration occurred in June 2022, when the above symptoms in the external genital area occurred.

During the conversation, the patient described her complaints and also reported that she had never before shown sexual interest, had no sexual desire, rated her libido at 0, and had never before experienced orgasm. She reported isolated cases of erotic dreams, after which

a feeling of guilt was present. She was scared of men, afraid of being a "toy for intimate pleasures". During the onset of these painful sensations, she first tried masturbating, hoping that this would reduce the intensity of the feeling of tension and pain, but the orgasms she achieved brought no relief: "It only got worse afterwards, and I also felt a feeling of guilt".

In general, the patient has a negative attitude towards the manifestation of her sexuality, explaining it by religious family traditions; she also stated that she wished this sphere in her life did not exist at all, and that she does not plan to have sexual relations in the future and to build relationships.

Based on the results of the consultation, the patient was recommended to see a neurologist and a gynecologist in order to exclude concomitant pathology. At the same time, pregabalin was prescribed at a dose of 150 mg/day, this dose of the drug allowed to almost completely eliminate the symptoms of the given disorder, however, after 3 days the symptoms returned by about 60–70 % according to the patient's subjective sensations. In this regard, the dose of pregabalin was increased to 225 mg/day, which reduced given symptoms by 50 % and normalized sleep and got rid of spontaneous orgasms. However, painful sensations localized in the clitoral area continued to maladjust the patient. Against the background of a decrease in these painful sensations, symptoms of the affective sphere began to actualize in the form of an increase in anxiety and depression. During the examination by a neurologist and an magnetic resonance imaging (MRI) scan, coccyx subluxation and spondylarthrosis were revealed. The diagnosis was determined: coccydynia, anxious-depressive syndrome. The therapy with meloxicam and diclofenac was prescribed, which had no effect on the symptoms in the genital area.

An examination by a gynecologist revealed no organic pathology of the genitals, but urinalysis revealed conditionally pathogenic flora, for which antibacterial therapy was prescribed, which neither lead to improvement in symptoms, nor caused thrush, followed by an increase in discomfort in the genital area. Therapy with antifungal agents was prescribed, after completion of which there were no pathological changes in the genital area.

In turn, the patient underwent hormone therapy by a gynecologist for polycystic ovarian syndrome, and for a year she took ethinylestradiol 0.03 mg/day and levonorgestrel 0.05 mg/day.

As a result, in a month and a half, after excluding the connection of these symptoms with organic pathology, considering the absence of sufficient effect of pregabalin and the increase of affective symptoms, it was decided to add escitalopram to therapy. Escitalopram was prescribed at a regimen dose of 5 mg/day with an increase to 15 mg/day, the pregabalin dose was reduced to 75 mg/day. As a result, during the next 2 weeks, the genital symptoms almost disappeared, the patient considered them to be 10 % of the initial ones, which did not affect her vital activity, but there was a significant increase in depressive symptoms.

Considering the patient's history, escitalopram and pregabalin were replaced by sertraline at a dose of 100 mg/day and lamotrigine 50 mg/day, and the patient also began using an ointment with a topical anesthetic as needed during episodes of increased symptoms. After 2 weeks, the patient reported stable gradual improvement in her emotional state and complete absence of genital symptoms.

Mental state. The patient is well groomed outwardly. The facial expression is weakly modulated, sad. She is available for productive contact, answers questions in a detailed way, and is fixed on her painful experiences. The voice is strained. While describing the symptoms in the genital area, she is shy, feels uncomfortable, her face turns red, tries not to call the organs by their names, uses expressions such as: "there is tension there", "it hurts in that area". Mood is depressed, anxious, tearfulness is present. Attention is unsteady, attention span is reduced. Thinking consistently. Memory and intellect are not impaired, the knowledge stock corresponds to the education received. Reports the presence of suicidal thoughts, which she does not intend to implement. Critically assesses her condition, asks for help.

Objective data. At the beginning of treatment (June 2022), the patient completed interviews on the GAD-7 and PHQ-9 scales with a PHQ-9 score of 13 and a GAD-7 score of 19. It was also proposed to rate the symptoms on a scale of 0 to 10 (VAS). Results: genital pain – 9, sleep disturbance – 8, sleep depth disturbance – 4, sleep quality – 6, physical condition limited daily activities – 10, emotional condition limited daily activities – 6.

The patient was examined (Aug. 31, 2022) with scales with a PHQ-9 score of 24 and a GAD-7 score of 15. It was also proposed to evaluate the VAS scale for genital pain – 3, sleep disturbance – 3, sleep depth disturbance – 3, sleep quality – 8, physical condition limited daily activities – 6, emotional state limited daily activities – 6.

MRI: coccyx subluxation and spondylarthrosis.

A blood test for the level of thyroid hormones: indicators are within the normal range.

Related specialists. Neurologist: Coccydynia; Anxiety-depressive syndrome. Gynecologist: Polycystic ovarian syndrome.

Diagnosis: Recurrent depressive disorder, current episode of moderate severity with somatic symptoms. F33.11

Associated diagnosis: Polycystic ovarian syndrome; Coccydynia.

Discussion

The PGAD studies point to the need to exclude organic pathology of the external genitalia [8,9]. Our patient was under constant observation by a gynecologist for polycystic ovarian syndrome and the gynecologist also excluded pathology of the external genitalia.

Some studies indicate a correlation between the onset of PGAD symptoms and taking or stopping antidepressants including SSRIs [10,11]. In the case we described, the correlation between discontinuation of an antidepressant and the beginning of symptoms and their disappearance when the drugs of this class are re-prescribed is clearly observed.

According to the available data, the occurrence of PGAD symptoms is significantly influenced by the patient's sexual beliefs, attitudes toward their sexual feelings and manifestations, and a sexual trauma in the

anamnesis [12,13]. In this case, the patient had developed under strict Christian beliefs in terms of sexuality, any of her own sexual manifestations led to feelings of guilt and shame. In addition, there was an episode of attempted physical abuse during adolescence, which significantly maladjusted the patient.

Some studies point to the relationship between hyperthyroidism and sexual dysfunction in men and women [9]. Our patient was examined for thyroid hormone levels, which were within normal range.

The role of inflammatory processes (urethritis, cystitis, vaginitis, thrush) in the formation and development of PGAD symptoms is pointed out [14]. In the case of this patient, symptoms of cystitis and thrush were observed, which significantly aggravated the clinical manifestations of PGAD; however, after recovery from the inflammatory processes, PGAD symptoms decreased, but remained at a rather high level.

There is an evidence of a correlation between symptoms of PGAD and pathology of musculoskeletal system, especially the lumbosacral spine [15–18]. The patient was examined by MRI and consulted by a neurologist and a subluxation of the coccyx was detected, and treatment with NSAIDs was prescribed, which had no effect on the symptoms of PGAD.

As a differential diagnosis, we considered a variant of psychalgia associated with withdrawal of antidepressants, but in the patient the symptoms of undesirable arousal were in the foreground, and the pain component was additional and manifested in a long-term persistent arousal state.

Conclusions

1. The presented case of the PGAD disorder is quite rare in medical practice and its clinical picture turned out to be typical in comparison with the results of other researchers.

2. The peculiarity of the described clinical case of PGAD is in possible association of symptoms with withdrawal of SSRIs (escitalopram).

3. The described case of PGAD confirms the data about high suicidal risk in this disorder.

4. The patient's treatment requires inclusion of specialists from different branches of medicine (neurology, gynecology and psychiatry) to rule out organic pathology and use of a multidisciplinary approach.

5. SSRIs (sertraline) and mood stabilizers (lamotrigine) were effective in the treatment of this case of PGAD disorder.

Prospects for further research. Detection and analysis of new cases of PGAD, development of standards of diagnostics, treatment and care, taking into account the need for a multidisciplinary approach to the management of this contingent of persons. Practitioners prescribing SSRIs should consider the possible risks of PGAD in patients.

Funding
This study was conducted in the framework of the scientific research work of Dnipro State Medical University "Development of modern technologies of differentiated psychiatric care for people with non-psychotic mental disorders and their relatives and mental health professionals", state registration No. 0121U114305 (2022–2026).

Conflicts of interest: authors have no conflict of interest to declare.
Конфлікт інтересів: відсутній.

Надійшла до редакції / Received: 22.12.2022
Після доопрацювання / Revised: 24.02.2023
Прийнято до друку / Accepted: 02.03.2023

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