Persistent Genital Arousal Disorder Treated with Duloxetine: A Case Report

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SUMMARY

Persistent Genital Arousal Disorder is characterized with unwanted, uncontrollable and persistent genital arousal symptoms that occur spontaneously in the absence of simultaneous sexual fantasy, sexual desire or sexual stimulation. The condition may last for hours or days. Patients often find it difficult to share this condition with their health care providers because they are afraid of being diagnosed with hypersexuality and they often get different psychiatric diagnoses such as Obsessive Compulsive Disorder and Major Depressive Disorder. Therefore, little is known about the real prevalence, pathophysiology or etiology of Persistent Genital Arousal Disorder. In addition, since there is no study conducted in this field, our information in this area is limited to case reports. Although there is no consensus about the treatment of Persistent Genital Arousal Disorder in the psychiatric literature, there are some case reports about the use of pregabaline, clomipramine, duloxetine, clonazepam, varenicline, olanzapine, risperidone in addition to the case reports on treatment with hypnotherapy, pelvic floor physiotherapy and electroconvulsive therapy (ECT). In this case report, we aimed to present the detailed description of a successful treatment procedure with duloxetine in a forty two years old female patient diagnosed with Persistent Genital Arousal Disorder. She had been using various antidepressants, antipsychotics, anxiolytics and mood stabilizers for sixteen years with different psychiatric misdiagnoses like Bipolar Disorder, Obsessive Compulsive Disorder, Anxiety Disorder and Major Depressive Disorder and yet, had not shared her symptoms of genital arousal with any psychiatrist previously.

Keywords: Persistent genital arousal disorder, duloxetine, treatment

INTRODUCTION

Although Persistent Genital Arousal Disorder (PGAD) is not a common disorder, it is a severe clinical condition that is closely associated with feelings of shame, social isolation and suicidal ideation (Leiblum et al. 2007). It was first described by Leiblum and Nathan (2001). PGAD occurs with non-sexual stimuli or without any stimuli and involves genital arousal symptoms lasting hours or days and not fully resolving alone. Physiological genital arousal responses are experienced frequently independent of subjective sexual desire or wishes; they occur in uninvited, intrusive or unwanted forms and cause problems. This situation, which is not resolved by normal orgasmic experiences, may subside with multiple orgasms over hours or days.

True prevalence of PGAD should be higher than what has been reported, this being possibly due to women with PGAD complaints fearing being ashamed of being diagnosed with hypersexuality and choosing not to share their complaints with health care professionals. There is very little known about the pathophysiology or etiology of PGAD. It may be associated with psychological-sourced pathophysiology including depression and anxiety, but may also be associated with pathophysiological processes with vascular, neurological or pharmacological basis (Leiblum et al. 2007, Thorne and Stuckey 2008, Komisaruk and Lee 2012). As the cause is not fully determined, a standard treatment algorithm is not available. Although there is not a consensus, there are case reports about the use of pregabalin, duloxetine,

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clonazepam, varenicline, olanzapine and risperidone, along with hypnotherapy, physiotherapy and ECT in the medical literature (Jakovich et al. 2016).

This report aims to review the case of a female PGAD patient who had been using a variety of antidepressants, antipsychotics, anxiolytics and mood regulation medications for different diagnoses like bipolar affective disorder, obsessive compulsive disorder (OCD), anxiety disorder and major depressive disorder (MDD) and her treatment process with duloxetine.

CASE

The 42-year old single, female, university graduate, primary school teacher, who had been unable to work for the last 5 years and lived with her family, was admitted to our outpatient clinics with depressive complaints and ongoing suicidal ideations.

The patient had been monitored from the age of twenty-six years with a variety of diagnoses. Mood status examination found that the patient had reduced self-care, was depressed and tense and displayed a defensive attitude. Affect and mood were depressed. Thoughts about the future were pessimistic, tainted with guilt and suicidal ideation. Sleep and appetite were reduced.

During the first days at the inpatient clinics, the patient was vicious and irritable and was observed to sleep very little at night. Interviews with the patient revealed that the patient had intrusive genital arousal which she had never shared with any psychiatrics before. In addition to genital arousal, the patient described clitoral pain. She reported hours of masturbation and multiple orgasmic experiences to resolve the genital arousal. She stated genital arousal occurred on its own especially in the evenings or nights without any previous feeling of sexual desire. The patient stated she had performed uninterrupted masturbation in the previous year from 23:00 PM until 04:00 A.M. every night, followed by intense thoughts about guilt, regret and sinfulness. She remained for hours in the bathroom for compulsive ritual ablutions due to obsessions of having soiled her body by masturbating. At the time of her admission as an inpatient she had masturbated for nearly 7-8 hours every day over the previous 10 days including remaining in her room for 2-3 days for continuous masturbation.

Her sexual history included first menstruation at 12 years of age and starting to masturbate at 11-12 years. In that period, she created sexual fantasies before masturbation and then did not feel guilt or sinfulness. The patient, although single, previously had a boyfriend. However, she had not experienced sexual intercourse with vaginal penetration due to religious beliefs. She did not have a history of sexual harassment.

Excessive genital arousal complaints occurred in the last years at university with excessive masturbation of 3-4 hours nearly every day which was not accompanied by an increase in sexual desire or impulses. Due to religious beliefs she had intense feelings of guilt and sinfulness linked to masturbation and spent hours in the bathroom performing repeated ritual ablutions for feeling dirty. The patient, therefore suffered from insomnia at night when she spent hours chatting on online dating sites until the morning. As she did not share her complaints with anyone, a psychiatrist attributed her lack of sleep to irritability and hypomania and she was started on valproate (500 mg/day) with Bipolar Affective Disorder type 2 diagnosis. Without clear benefit from treatment, the patient stated she used medications like lithium, paroxetine, fluvoxamine, clomipramine, imipramine, clonazepam, amisulpride and zuclopenthixol at doses she could not remember later at psychiatric follow-up. When she was thirty-four years old, she was admitted to a university hospital with OCD and MDD diagnoses due to depression, anhedonia, feeling troubled, insomnia, thoughts about guilt and sinfulness and spending hours in the bathroom for treatment. The patient did not mention her masturbation complaints to the clinician at the time and could not masturbate due to using a shared bathroom so rejected treatment at the end of the first week and discharged herself. She stated she did not benefit from the clomipramine treatment.

With the increase in excessive genital arousal and masturbation behaviour, the patient had not been able for work for the last 5 years and had irregularly used a variety of medications which she could not remember. The last medications used by the patient were quetiapine 200 mg/day and sertraline 200 mg/day, without benefit. In the absence of manic and hypomanic periods wor diferential diagnosis of Bipolar Affective Disorder, the patient was considered to be hypomanic by a psychiatrist while a university student for being excessively irritable due to inability to sleep and chatting on online dating sites until morning and. at night. The patient stated that she did not share with her doctor her complaint of not being able to sleep because of excessive desire for masturbations due to enital arousal. The patient and her family stated she had never had any of the other accompanying symptoms like energy increases, elevated self-esteem, or excessive spending. Interviews indicated that she performed repeated ritual ablutions due to obsessions about guilt and feeling dirty as a result of excessive masturbation when she did not leave the bathroom for hours. She was diagnosed by another physician with OCD on the basis of these symptoms. However, the obsessive and compulsive symptoms only occurred following excessive masturbation due to PGAD and were associated with feelings of guilt linked to the patient's religious beliefs. There was not any symptom of obsession or compulsion such as control, symmetry or counting.

Gynecological examination and accompanying pelvic and transvaginal USG tests with the aim of assessing the presence of a clitoral mass did not encounter any pathologic findings.

The patient's periods were regular and normal, with FSH, LH, estrogen and testosterone values within normal limits and endocrinology consultation excluded hormonal causes. Cranial, pelvic and sacral MRI were requested to exclude mechanical causes such as central and peripheral neurological anomalies, pelvic congestion and pudental nerve compression and pathological findings including Tarlov cyst were not observed. Neurology consultation did not identify restless legs syndrome. EEG taken to exclude possible epileptic foci and EMG to exclude small fiber sensory neuropathy were within normal limits.

At the patient's admission, her scores were 41 on the Hamilton Depression Rating Scale (HAM-D-Akdemir et al. 1996), 45 on the Beck Depression Inventory (BDI-Hisli 1988), 48 on the Hamilton Anxiety Rating Scale (HAM-A-Yazıcıet al. 1998) and 53 on the Beck Anxiety inventory (BAI- Ulusoy 1993). Initially clitoral pain severity was assessed with the 1-10-point horizontal visual analog scale (VAS) and her VAS points were calculated to be 9 (McCormack et al. 1988). In light of these evaluations the preliminary diagnosis was PGAD accompanying major depressive disorder. Sertraline (200 mg/day) and quetiapine (200 mg/day) started at her admission were reduced and stopped. In addition to diazepam (5 mg/day) duloxetine (30 mg) was added to clear depressive complaints and the described clitoral pain. Two weeks later duloxetine was increased to 60 mg/day; and diazepam was stopped at the end of the first month. Upon learning that the patient continued to masturbate when staying in a single room, she was given the homework to monitor daily incidences of masturbation on a table. Psychotraining related to PGAD was given and attempts were made to reduce feelings of guilt. After the first month of treatment, genital arousal symptoms had regressed by 80%, and the total masturbation duration of 7-8 hours observed at hospital admission was reduced to an average of half an hour. Also, obsessive compulsive complaints of the patient resolved, and the clitoral pain was given 1-2 VAS points. At the end of forty-five days after admission, the patient was discharged with only 60 mg/day duloxetine treatment and scores of 9, 10, 11and 11 on, respectively, HAM-D, BDI, HAM-A and BAI. Masturbation behaviour was reduced to 15 minutes. In the third month of treatment, genital arousal and pain complaints had completely resolved with no masturbation behaviour disturbing the patient during the day. With daily functioning returned to normal, the patient did not describe any depressive or obsessive-compulsive complaints.

DISCUSSION

This case presented the five symptom clusters described for PGAD (Leiblum and Nathan 2001). Though not often encountered in women, it is necessary to exclude hypersexuality displayed with frequent masturbation, persistent and intrusive sexual fantasies or thoughts and increased frequency of sexual intercourse (Kafka 2010). Incontrast to hypersexuality, PGAD is defined by the presence of genital arousal without sexual desire, as in this case.

Women with PGAD diagnosis are reported to have severe disturbance of mental health in addition to difficulties in maintaining sexual functioning and daily activities. Leiblum et al. (2007) compared women with full diagnostic criteria for PGAD with those with only some. Among those fulfilling all criteria, there were more depression, anxiety, panic attacks, negative affect related to physical sensations and genital sensations identified. Although our patient was a teacher, she had not worked for the previous 5 years, spending a significant part of her daily life masturbating, feeling troubled and getting irritated when prevented from masturbating. With the increase in severity of depressive complaints in the recent period, suicidal ideation was added. Religious beliefs, feelings of guilt and dirtiness due to excessive masturbation led the patient to repeatedly perform ritual ablutions and she was previously followed up for OCD diagnosis. Similarly, a series of PGAD cases monitored for mistaken OCD diagnosis due to frequent repeated ritual ablutions after masturbation was reported previously in Turkey (Yıldırım et al. 2012).

The pathophysiology and aetiology of PGAD is not fully known. In our case there was not any causative factor or medical situation that could explain the complaints. As there is no standard treatment algorithm, detailed psychiatric and physical disease history and examination, along with treatment choices for accompanying diagnoses indicate the route taken. Our case initially began diazepam treatment due to insomnia and restlessness complaints. However, with clarification of the diagnosis, duloxetine was added to her treatment for depression and anxiety symptoms accompanying clitoral pain and later the dose was increased to 60 mg. Her well-being observed at the end of the first month led to cessation of diazepam and continuation of duloxetine treatment alone.

Duloxetine is a serotonin and noradrenalin reuptake inhibitor used for MDD, generalized anxiety disorder, pain associated with diabetic neuropathy, fibromyalgia and chronic musculo-skeletal pain (Jost and Marsalek 2004). Our reason for choosing this medication is that it has analgesic properties in addition to antidepressant and anxiolytic effects. Some authors (Waldinger et al. 2009) consider PGAD may be the possible result of a neuropathy affecting the small fibers of the pudental nerve. The efficacy of duloxetine here may be that it increases the tonus of inhibitory pain fibers descending from the brain and spinal cord (Bellingham and Peng 2010). This inhibition is similar to the effect defined in diabetic neuropathy and fibromyalgia. Additionally, it is possible duloxetine has an inhibitory effect on sacral and/or thoracolumbar neurons excited by genital sexual arousal and orgasm (Jost and Marsalek 2004). In similarity to our case,

there are two case reports of treatment with duloxetine in the literature, but the specific mechanism of effect of duloxetine for this disorder is still speculative (de Magalhães and Kumar 2015, Philippsohn and Kruger 2012).

PGAD is still a topic that is insufficiently researched. The low prevalence reported may be linked to a lack of sufficient questioning and diagnosis of the disease due to lack of awareness of this topic among healthcare workers. Presence of PGAD symptoms should be questioned in detail as a component of comprehensive sexual and reproductive history by family doctors, gynecologists, urologists and algologists, in addition to psychiatrists. There is need for structured research about treatment of this disorder in the long-term.

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