CLINICAL PSYCHOLOGY & NEUROPSYCHOLOGY | CASE REPORT

Treatment of persistent genital arousal disorder: Single case study
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Abstract: This abstract gives the case description of a married woman in her 30s who suffered from persistent genital arousal disorder for a decade. This paper reviews the efficacy of mindfulness-based cognitive therapy (MBCT) in the management of her disorder. The client had been reported as experiencing persistent genital arousal, vaginal engorgement, accompanied by feelings of distress, irritability and psychosocial impairment. She had a history of marital discord with her husband as well and never get sexual satisfaction. Besides, she had also suffered from dysthymia for the last eight years. She was prescribed antidepressants for dysthymia and local anesthetic ointment for genital irritability. She appeared normal in her medical evaluations. She reported mild relief in her genital arousal and dysthymia in response to psychiatric medication. She was treated with MBCT for a period of six months. Six months periodical follow-up reported a moderate improvement in symptoms of dysthymia, distress, and persistent genital arousal. She had been advised to see her therapist at regular intervals for periodic evaluation and administration of MBCT. She has been instructed to practice mindfulness exercises on her home regularly to keep the effects intact.

Subjects: Social Sciences; Behavioral Sciences; Mental Health

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PUBLIC INTEREST STATEMENT
Persistent Genital Arousal Disorder (PGAD) is an emerging sexual disorder that occurs infrequently and affects both men and women, although, more women have been found to develop this condition. The researchers’ understanding of the causes of this emerging condition is still evolving. Newer treatments have been proposed by various theorists. The present article is a clinical case study of a 38 years old Muslim married woman named Saima treated effectively with mindfulness-based cognitive behavior therapy (MBCT). Saima presented with complaints of persistent genital arousal in the absence of subjective arousal accompanied by personal distress for over a decade. She was treated effectively with the 6 months administration of MBCT involving mindfulness in addition to cognitive behavioral techniques. The MBCT was also found effective when assessed post treatment 6 months later. Saima was advised to practice mindfulness regularly to keep the effects intact.
Keywords: persistent genital arousal disorder; depression; sexual & relationship; difficulties; mindfulness; cognitive behavior therapy

1. Introduction
The persistent genital arousal disorder (PGAD) is a rare condition mostly affecting women causing them a lot of distress. The term was first used by Modell in 1989 (Modell, 1989) and was first defined by Leiblum and Nathan as Persistent Sexual Arousal Syndrome (PSAS). Later, they renamed it as PGAD to emphasize its genital rather sexual nature (Leiblum & Nathan, 2001). This refers to an uncomfortable condition characterized by non sexual, persistent genital arousal and engorgement of clitoris and labia produced mainly in the absence of sexual stimuli. The sufferer typically experiences clitoral pain or pressure and vaginal engorgement, sometimes spontaneous orgasms. This condition is not relieved by orgasms and causes a lot of distress and dysfunction in the life of the sufferer. There has been shame, guilt and hesitation regarding seeking help about PGAD symptoms (Aswath et al., 2016).

The workable definition of PGAD was first developed by Leiblum in 2005 (Leiblum et al., 2005) and was described in full detail later (Leiblum & Goldmeier, 2008).

Now, for the diagnosis of PGAD, the following conditions should have been met:

1. Symptoms characterizing sexual arousal that is persistent and does not end on its own
2. Symptoms of sexual arousal usually do not subside with orgasm but continue for hours or days and remit with multiple orgasms over a period of hours and days
3. The symptoms of sexual arousal are developed and maintained in the absence of real subjective sexual desire or feelings and there is weak evidence of genital arousal (lubrication) upon examination
4. The development of symptoms of sexual arousal is not usually triggered by sexual stimuli
5. The symptoms of sexual arousal are experienced as causing distress and impairment in psychosocial functioning
6. The symptoms are experienced as intrusive and unwanted
7. The symptoms become aggravated by certain circumstances like sitting, riding, driving or nervousness (Basson et al., 2010).

The exact prevalence of this rare condition is not known as the most cases of PGAD are described in literature in the form of case studies. In the past, only women were thought to be affected by this condition but now some men have been found to suffer from this rare condition as well (Kamatchi & Ashley-Smith, 2013; Kruger & Hartmann, 2016; Waldinger et al., 2011). Women of all age groups can be affected by PGAD (Goldmeier et al., 2009). However, the PGAD is prevalent in 25–51 years old women and 38–74 years old men (Kamatchi & Ashley-Smith, 2013; Leiblum & Goldmeier, 2008; Pink et al., 2014; Reading & Will, 1997; Waldinger, Van Gils, et al., 2009; Waldinger et al., 2011). A recent study estimated the prevalence of PGAD in two North American non-clinical samples to be 6.8% to 18.8% in women (Jackowich & Pukall, 2020). In these samples, the most frequent symptom of PGAD was sexual arousal in the absence of sexual stimuli. Majority of the cases occurred in menopausal and peri menopausal phases. Majority of females who
experience PGAD have in a long-term relationship as well (Leiblum & Chivers, 2007; Waldinger, Van Gils, et al., 2009). The pathophysiology and exact etiology of PGAD remain unknown (Broto et al., 2010; Goldmeier et al., 2009; Waldinger et al., 2011).

Etiological factors are not specific and represent the features of case studies available in the literature. A total of 13 case studies of PGAD point towards the occurrence of anxiety, depression or obsessive compulsive features and chronic fatigue as possible etiologies of PGAD (Goldmeier & Leiblum, 2008; Goldstein et al., 2006; Leiblum & Goldmeier, 2008; Mahoney & Zarate, 2007; Yero et al., 2006). Sexual abuse and sex-related emotional problems such as concerns about body image and sexual incompatibility between partners have also been found to be present in cases of PGAD (Waldinger, Venema, et al., 2009). Some studies pointed toward the use of Selective Serotonin Reuptake Inhibitors (SSRI) such as fluoxetine as the etiology of PGAD (Leiblum & Goldmeier, 2008). Though, some other studies indicated otherwise and reported the discontinuation of SSRI as the etiology of PGAD (Freed, 2005).

Leiblum and Chivers (2007) explain why mental health conditions comorbid with PGAD. According to them, there may be discordance between women's psychological state and their physiological arousal. Anxiety states lead to experiences of genital arousal in some women, e.g., engorgement of genitals (Bradford & Meston, 2006) as well as the increase in attention to threatening or unpleasant sensations (Lang et al., 2000). Leiblum and Chivers argue that genital arousal sensations are recognized as negative possibly because of past experiences. This negative experience leads to more anxiety which leads to further genital arousal and cognitive distortion (Leiblum et al., 2007). As depression often overlaps with anxiety and chronic fatigue, therefore these conditions are comorbid with PGAD. Thus, being worried and anxious about having persistent arousal actually maintains symptoms of PGAD (Leiblum & Chivers, 2007).

On the other hand, Persistent Genital Arousal Disorder can be developed in response to clitoral priapism. But this condition causes more pain than symptoms of PGAD (Goldstein et al., 2006). Other conditions associated with PGAD include pelvic vascular abnormalities, and damage or irritation of pudendal nerve or small fiber sensory neuropathy (Waldinger & Schweitzer, 2009; Waldinger et al., 2011; Waldinger, Venema, et al., 2009). In some patients, the sitting position aggravates the complaints of PGAD while walking around relieves the symptoms (Waldinger & Schweitzer, 2009). The symptoms appear to aggravate in evening or night hours (Facelle et al., 2013).

Resultantly, no single treatment approach is suggested for sure to bring improvement in clinical picture (S. R. Leiblum, 2003). The treatment is usually symptomatic (S. Leiblum, 2006). Benzodiazepines, tricyclic antidepressants (TCA), selective serotonin reuptake inhibitors (SSRI) and anesthetizing gels are often prescribed for the anxiety, depression and compulsive behavior associated with PGAD (Bell et al., 2007; Hiller & Hekster, 2007; Korda et al., 2009; Waldinger & Schweitzer, 2009; Waldinger, Venema, et al., 2009). GABAergic drugs like gabapentin and pregabalin have been prescribed in some cases of PGAD (Philippsohn & Kruger, 2012). Psychotherapy including cognitive behavior therapy and couple therapy has been found useful in cases of PGAD (Hiller & Hekster, 2007; Lofrisco, 2011).

The PGAD's influence on a woman's health and daily functioning is enormous. Recent studies indicated that the symptoms of PGAD were found to be associated with mental health concerns such as increased stress, anxiety, and depression as compared to symptom free women (Carvalho et al., 2013; Jackowich et al., 2020; Squibb et al., 2019). The women with PGAD had been found to show poorer psychosocial functioning such as related to daily social and work activities (Jackowich et al., 2018). The PGAD was also found to affect the relationship and sexual satisfaction of women (Squibb et al., 2019).
Given the impact of PGAD on women lives, there is scarcity of studies documenting the efficacy of psychotherapeutic management of PGAD (Jackowich et al., 2016). Although, studies recommended the use of psychotherapies for the treatment of PGAD (Facelle et al., 2013; Goldmeier et al., 2014) but only one study reported the effects of couple therapy using cognitive behavioral techniques (Hiller & Hekster, 2007). The present article was meant to fill this gap in literature on psychotherapeutic management of PGAD in women. The present article describes the case of a married woman named Saima (fictitious name), aged 38 years who had been treated with mindfulness-based cognitive psychotherapy (MBCT) besides psychiatric medication and local analgesics. The present article aimed to report the efficacy of mindfulness-based cognitive psychotherapy (MBCT) in relieving symptoms of PGAD in Saima.

2. Methodology
The present case is a clinical case study of exploratory nature which aimed to explore the effectiveness of MBCT in treating the symptoms of PGAD in middle-aged woman. The PGAD was diagnosed in Saima based on the diagnostic criteria given by Leiblum and Nathan (2001). The MBCT included components like psychosexual education, cognitive restructuring, stress management, present moment non-judgmental acceptance, and self-compassion. The client did not report any side effect of MBCT. The improvement in symptoms was assessed based on the similar diagnostic criteria and was described with the help of relevant theories. The diagnostic criteria were used to assess improvement in symptom to avoid the therapist biasness. The written informed consent was obtained from Saima before enrolling her for the therapy. The written consent was obtained from Saima for reporting of case for publication.

3. Case study
Saima is a married heterosexual woman aged 38 years who had been suffering from symptoms of persistent genital arousal, vaginal engorgement and genital pain for a decade. She got married 13 years ago and started having heterosexual relations with her husband. She had not been satisfied sexually with her heterosexual relationship with her husband during this entire period. She did talk to her husband about her sexual dissatisfaction but the things could not get better. Apart from sexual dissatisfaction, she started developing relationship difficulties with her husband after few years of marriage. These relationship difficulties and emotional coldness further aggravated the experience of symptoms of sexual dissatisfaction. On the other hand, she had been suffering from brief, persistent episodes of sad and low mood combined with somatic features including episodes of binge eating and constipation for the last eight years calling for a diagnosis of mood disturbance. She occasionally experienced sudden and prolonged bouts of arousal in her genitals in the absence of sexual stimuli before marriage.

She had reported doing masturbation to relieve her of uncomfortable vaginal sensations. But she was not always successful in controlling her unstoppable genital arousal before marriage. Her symptoms of persistent genital arousal started to increase in frequency and intensity after a few years of her marriage. The symptoms of genital arousal occurred at least twice a week, ranging from two hours to two days. The symptoms aggravated in situating position or by psychological nervousness. She tried different non-medicinal strategies to get relief like cold shower, distraction, masturbation and increased sexual intercourse. But these strategies could not make her symptom free and brought little to no relief.

She had contacted general physician, gynecologist, neurologist and a psychiatrist before coming for psychotherapy. She had been tested for clinical picture of blood, urinalysis, hormonal assays, thyroid function, liver function, kidney function and diabetes. All her blood and urine tests appeared normal. She had been examined for gynecological and neurological abnormalities by the concerned specialists but nothing pathological could be find. All her symptoms led to the
diagnosis of PGAD and persistent depressive disorder (dysthymia) as per diagnostic criteria of DSM-5 (American Psychiatric Association, 2013) and Leiblum and Nathan (2001) respectively.

4. Background information & formulation of the case
Saima had been gone through her childhood and adolescence as a traditional Muslim girl. She was the eldest of four siblings including three sisters and one brother. Her father died during her childhood. All her developmental milestones were normal. She reached menarche at the age of 12 years. Generally, she used to be a shy and coy girl in her adolescence, she did not have relationship with opposite sex during adolescence. She only dreamed of having a perfect partner for her in the form of husband. She got married at the age of 25 years with the permission of her mother. Saima’s sexual relations with her husband were never satisfied. She could not satisfy her need of emotional intimacy and love with her husband. She termed the sexual relations with her husband as mechanical only. Interestingly, she considered fulfillment of sexual activity a kind of duty to the husband because of her traditional upbringing. She used to complain about her dissatisfaction to her husband but he never listens to her seriously. Soon after marriage, her relationship with her husband witnessed bickering, complaints, and dissatisfaction. She had two kids, one boy and one girl. She gave birth to both kids through Cesarean section. Saima’s general health was good overall. She did not suffer from any major or terminal illness. She also had no history of any major terminal or psychiatric illness in the family.

Saima’s complaints of persistent genital arousal in the absence of subjective sexual feelings present for hours and days, not remit by orgasm, and aggravated by sitting position started worsening after few years of marriage. Although, she occasionally experienced bouts of persistent genital arousal before marriage but those episodes were not so problematic for her. After few years of her marriage, her complaints of persistent genital arousal started occurring more frequently and intensely. Concurrently, she started having troubles in her marital relations with the husband and started developing relationship difficulties. At the same time, she started experiencing episodes of low and sad mood accompanied by somatic features. Her persistent arousal may in part be attributed to the use of SSRI and in part attributed to the dissatisfied sexual relations and relationship difficulties with her husband, though, the possibility of SSRI as an etiology is weak as she used to have occasional episodes of persistent arousal well before the start of SSRI. Keeping in view these kinds of psychological and emotional pathology, mindfulness-based cognitive psychotherapy was suggested to her to make herself manage psychological and emotional problems.

5. Treatment
She had been prescribed several medicines for persistent genital arousal, pain, mental anguish and mood swings. She was prescribed with alprazolam to make herself calm and pain free but she did not get better. She was prescribed nortriptyline for episodes of dysthymia and genital pain followed by escitalopram which brought moderate relief in her mood disturbance. She used local anesthetic ointment like lignocaine or lidocaine to make her genital area pain free which brought some relief in genital pain. She was treated with techniques of behavioral, cognitive and mindfulness psychotherapy. Couple therapy could not be initiated with these approaches as her husband failed to come for therapy. The cognitive restructuring of her beliefs regarding sexuality like “vaginal sex is the only way of getting sexual satisfaction”; “sex is a kind of duty to your husband”; “I can not make my husband happy sexually”; and “I will not be able to satisfy myself sexually” was done by the therapist. She was taught muscular relaxation combined with imagery, distraction and mindfulness over a period of 24 weeks with a ratio of two sessions per week. This led her to experience reduction in her symptoms of genital arousal both in frequency and intensity. The genital arousal occurred once a week ranging from 1 to 1.5 hours. The client reported a moderate relief in persistent genital arousal symptoms, sad mood and overall quality of mood over a six month period post treatment. The client was advised to see therapist regularly at six months
periods and practice mindfulness and stress management techniques regularly to keep her symptoms manageable. She uses mindfulness exercises currently.

6. Discussion
The present case study aimed at finding the efficacy of MBCT in amelioration of symptoms of PGAD in a middle-aged married woman. The client, Saima reported moderate improvement against all criteria of PGAD as compared to her pre-treatment condition when she reported all criteria as severe. Saima reported reduction in occurrence of sexual arousal from 2 hours–2 days twice weekly at pre-treatment to 1–1.5 hours once weekly post treatment. She reported reduced distress and improved mood during her social functioning on daily basis. She gained significant improvement in experience of genital arousal by sitting position.

Pharmacotherapy alone did not lead to elimination of symptoms of genital arousal. The psychotherapy especially mindfulness-based exercises found to bring moderate relief in her symptoms of persistent arousal, sad and low mood, and accompanied distress and irritability. The mindfulness had been found to bring improvement in sexual functioning of women in some past studies (Paterson et al., 2017), although, those studies found mindfulness effective in cases of sexual interest, and sexual arousal in women. No study in the past reported the efficacy of mindfulness-based cognitive therapy for reducing symptoms of PGAD, though, one study did report the efficacy of cognitive behavior therapy in couple therapy format for the treatment of PGAD in an old age woman (Hiller & Hekster, 2007). The techniques of stress management, self-compassion, present moment, non-judgmental acceptance of one’s condition combined with psychosexual education brought the moderate relief in sexual arousal, and mental irritability, and improved the daily functioning of Saima. The findings of this clinical case study are significant as adding to the existing literature of psychotherapeutic management of PGAD in women. This is the first clinical account of effectiveness of mindfulness-based cognitive techniques in improving symptoms of PGAD. The improvements in client’s symptoms and daily functioning were persistent at post-treatment which indicated the long-lasting effectiveness of MBCT.

7. Limitations & strengths
The main strength of this study is the effectiveness of MBCT in improving symptoms of PGAD and psychosocial functioning. The effectiveness of MBCT in treating symptoms of PGAD made MBCT a culture fair therapeutic technique. The results of the study could be quantified by the use of standardized tests to measure the effects of the therapy. The study lacked control group due to its case study design.

8. Conclusion & implications
It has been learnt that cognitive restructuring of beliefs regarding sexuality, mindfulness based exercises in combination with pharmacotherapy have been found to improve symptoms of PGAD, and reduce personal distress (Carvalho et al., 2013). We conclude that understanding of the PGAD is still evolving (Aswath et al., 2016). The patients with PGAD can live a better life if treated with combined therapeutic approach using MBCT.

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