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Case Report

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Persistent Genital Arousal in a Woman with Pudendal Neuralgia

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Abstract

Persistent genital arousal disorder has been identified as a condition of often unprovoked genital arousal and is not well understood. A number of potential causes have been proposed such the pudendal neuralgia. We present a woman with a history of depression who consulted for unwanted genital arousal. The patient experienced a partial improvement with cognitive behavior and antidepressant therapies. Symptoms were disappearing with rehabilitation for pudendal neuralgia. This case illustrates the need to recognize atypical persistent genital arousal, as these patients report unusual symptoms that should alert the physician.

Keywords: Sexuality, Pudendal neuralgia, Arousal

Introduction

The majority of women who are referred to psychiatrist are found to be complaining of loss of interest, orgasmic difficulties or some pain linked to penetrative sexual activity [1]. Much less common is the presenting problem of unwanted frequent sensations of sexual arousal.

We present a woman with a history of depression who consulted for unwanted genital arousal. The patient experienced a partial improvement with cognitive behavior and antidepressant therapies. Symptoms were disappearing with rehabilitation for pudendal neuralgia. The objective of this work is to study the links between these disorders through a clinical case.

Case report

Mrs. A., a 30-year old, married, Caucasian female was presented with uncontrolled and debilitating levels of sustained genital arousal at our psychiatric hospital on January 2016. Raised in a traditional Muslim family with strict principles, she commenced sexual activity at age 21 at which time she is married. She didn't work and had two daughters aged four and six. She did not report any history of sexual abuse or relationship problems and described a good marital and sexual relationship with her partner. At the age of 26, she was placed on paroxetine for management of depression. She also started to smoke cigarettes, two cigarettes per day for three months. After a year, she stopped treatment because she felt good.

She was too embarrassed to discuss her problem with anyone else, she reported that she has currently been experiencing symptoms of persistent and unwanted genital arousal in non-sexual circumstances. The frequency increased to 3-4 times a day. There was two year history of uncomfortable sensations of regular sexual arousal that could last for several hours at a time. Sometimes, this sensation seems like a pain wakes the patient at night. The location was restricted to the vagina with no other body parts involved. There was no actual desire to proceed with sexual activity when she had the sensation. Interestingly, intensity was described as variable, in part because of the time of day, and in part because of her mental state. The intensity worsened with stress, anxiety, fatigue, and in the afternoon. Driving a car and while sitting brought about the start of the sensations. She reported that employing some kind of mental distraction tended to reduce the intensity of the sensations. The gynecologist was considered it as a psychiatric disorder and didn't examine her. The neurologist didn't identify any physical problem and a further psychiatric opinion was sought. Endocrine assessments included prolactin, follicle stimulating hormone, luteinizing hormone, total oestradiol; progesterone, testosterone, sex hormone-binding globulin, free thyroxine, and thyroid Stimulating hormone were normal. In addition, vitamins and minerals were assessed. Folic acid, vitamin B12 and haptoglobin were also assessed and reference ranges were used according to the manufacturer manuals. Hormonal blood investigations were normal. A diagnosis of persistent genital arousal disorder (PGAD) was made. Antidepressant medication (amitriptyline 10mg) was tried and she had found this to be effective in reducing the sensations. She was also advised that psychological therapies might be a useful avenue to consider. Two sessions of cognitive therapy focused on Mrs. A. increasing her understanding of her anxiety about the arousal sensations. Information about anatomy and advises on how to wear clothes were provided to increase her confidence in her ability to control her own body. Third session focused on behavioral therapy such as relaxation exercises and appropriate distraction measures, such as the use of music. Mrs. A. arrived for the fourth session reporting that she felt much more relaxed about the arousal sensations and that she had been practicing the exercises she had learned at the previous sessions. However, she remained anxious and these sensations diminished gradually but

didn't disappear. She underwent electrophysiological recording: bulbo cavernosus muscle electromyography, measurement of the bulbocavernosus reflex latencies, somatosensory evoked potentials of pudendal nerve and pudendal nerve terminal motor latencies. Signs of denervation localized to the territory of the pudendal nerve were found. Pudendal neuralgia was diagnosed as comorbidity to persistent sexual arousal syndrome and rehabilitation was indicated.

However, the patient did not return for follow up. She e-mailed one week after the initial visit stating that her symptoms were markedly resolved. A follow-up phone call to the patient 12 months later confirmed that the resolution of her symptoms was complete. But, she cannot remember the number of rehabilitation's sessions.

Discussion

Medical symptoms that cannot be ascribed to organic illness can bring about different kinds of reactions in physicians who may need to reattribute this problem to other causes, including the possibility of a psychological aetiology [2]. In our case, the gynecologist and the neurologist didn't examine the patient because he cannot distinguish if it is psychiatric or physical problem or both.

In this case, physical examination was normal. In the literature, high rates of pelvic varicies and Tarlov cysts in women with PGAD have been described and discussed as possible contributors to PGAD [3]. In fact, PGAD can result from Tarlov cysts, which contain aberrant sensory nerve fibers and form on the genital sensory nerves where they abrade on the entrance to the sacrum, and/or herniated intervertebral disc-produced irritation of the roots of those genital sensory nerves as they course through the cauda equina within the spinal canal [3]. These conditions are not uncommon in women with PGAD and our patient did not undergo Magnetic Resonance Imaging of the sacral spine, which may account for some of this discrepancy.

The diagnosis criteria for PGAD include the presence of spontaneous and persistent genital arousal that causes distress, which patients often liken to pain or describe as painful. A number of the proposed etiologies include conditions associated with pain, such as central and peripheral neurological anomalies, mechanical impingements of the pudendal nerve, overactive bladder, and pelvic congestion [4, 5]. Pudendal neuralgia is a form of chronic neuropathic pain [6]. It was often misdiagnosed and inappropriately treated. Based on the description of her symptoms, it is believed that this patient meets all criteria for the diagnosis of persistent genital arousal disorder. She recognized and experienced persistent genital arousal feelings which were not related to excessive sexual desire.

Much of the available literature consists of case studies [5, 7]. There is still debate about the diagnosis itself and whether there is more than one disorder. In addition, there is no objective method of diagnosing persistent genital arousal disorder. In this case, the patient didn't distinguish between pain and persistent genital arousal and we diagnosed both persistent genital arousal disorder and pudendal neuralgia.

The patient has been improved partially with cognitive behavioral therapy and amitryptilline which is useful for neuropathic pain. Therefore, we conclude that his symptoms may be a result of interaction between physical and psychological factors. This suggests that PGAD could be a psychosomatic condition, which was already proposed as a cause for PGAD in women by Goldmeier and Leiblum [8]. In the presence of unusual clinical presentation, the diagnosis of pain due to pudendal nerve entrapment should be reconsidered, and a radiological examination should be performed. Peripheral nerve hypersensitivity and pelvic congestion are identified as two of the most likely etiologies; both can result in pelvic pain. Recently, it has been postulated that pudendal neuropathy was a key factor in Restless Genital Syndrome which consists of PGAD and/or restless legs and/or overactive bladder [8]. There seems to be overlap between persistent genital arousal and pelvic pain.

Our clinical experience is that combination of cognitive-behavioral therapy plus amitriptilline. Many classes of antidepressants may cause such interaction, including the tricyclic group and the selective serotonin reuptake inhibitors (SSRIs), which have an effect by increasing serotonergic tone and inhibiting dopamine-related activation of the sexual response [9]. The use of antidepressants can be helpful in women with PGAD, especially in those with obsessive traits, anxiety, or depression [9]. In fact, sexual arousal in women and men is under the control of both the autonomic and central nervous system [10]. Brain catecholamines like dopamine and noradrenaline play a causal role in the regulation of autonomic outflow and concomitant sexual desire and behavior [9]. Moreover, one potential cause of PGAD may be hyperactive dopamine release [11]. Antidepressant (Selective serotonin reuptake inhibitor, amitryptilline) could have a similar inhibitory action on dopamine release but with a high potential for sexual side-effects [11]. Also, surgical release of pudendal nerve entrapment has resulted in PGAD symptom improvement. Furthermore, chronic pudendal neuromodulation can be an effective treatment for decreasing frequency of PGAD symptoms [12].

Conclusion

This case illustrates the need to recognize atypical persistent genital arousal. All women should be asked about these symptoms, as they are often hesitant to discuss them despite the significant distress they cause. Specifically, women who present with pelvic pain, interstitial cystitis and other urinary pain, restless legs syndrome, and pudendal neuralgia should be asked about persistent genital arousal symptoms. Further study is required to understand the etiology of PGAD and how widespread and disabling this problem is, and to investigate approaches to management.

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