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CASE REPORT

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Is it Really Rare? Restless Genital Syndrome

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ABSTRACT

Restless genital syndrome (RGS) is a new definition in which awareness has begun to emerge in recent years. RGS is described as complex sensations of discomfort in the genital area, as itching, contraction, pain, and swelling in the absence of sexual stimulation. This condition, which may be quite uncomfortable for the patient, is hidden due to shame and not brought to the attention of physicians. Other problems that patients may encounter are that many physicians are inexperienced regarding such symptoms, that the family physician or the first physician does not know to which specialty they should refer patients with these symptoms, and that there is no clear consensus on treatment. Numerous etiologies, pharmacologic and non-pharmacologic treatment options have been discussed in the literature.

We discussed two female patients who were diagnosed as having RGS and benefited from treatment.

Keywords: Restless genital syndrome, persistent sexual arousal syndrome, restless leg syndrome

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INTRODUCTION

Restless genital syndrome (RGS) is a state of excessive and persistent genital and clitoral stimulation without a voluntary sense of sexual desire or activity. In the literature, the number of female patients is remarkable. Masturbation and sexual intercourse do not provide any real relief. Unlike hypersexuality, there is no real desire for sexual activity. This feeling may persist for hours or throughout the day and can be extremely uncomfortable for the patient. It is described in many different ways by patients. Some patients choose more ingenuous expressions such as tickling, discomfort, heartbeat, itching, and swelling feeling of vagina, whereas some patients may describe the feeling directly like an orgasm (1). The symptoms may increase and become unbearable at rest when the patient is lying or sitting for a long time. Although it is considered to be very rare, patients are more likely not to report it due to shame and embarrassment.

In 2001, Leiblum and Nathan described involuntary genital stimulation in patients with Parkinson's disease independent of sexual desire and called it persistent sexual arousal syndrome (PSAS) (2). In terms of pathogenesis, many theories have been proposed over the years: pudendal nerve neuropathy, pelvic varices, and vasocongestion (3, 4). The term persistent genital arousal disorder (PGAD) was proposed by Leiblum in 2007 because the disease is an involuntary genital sensation rather than sexual desire (5). In a study of 18 patients in 2009, similar symptoms were seen in 12 patients with a pre-existing diagnosis of restless leg syndrome (RLS), suggesting that dopaminergic mechanisms may be effective in the mechanism of the disease and RGS diagnostic name is suggested instead of PGAD (6).

Herein, we share two cases.

CASE 1

A 53-year-old female patient was admitted with symptoms of persistent discomfort in her vagina and legs, which had had for six months. Although she had no desire to have sex, she described having vaginal discomfort as pulsing, throbbing, itchy feelings, and sometimes discharge while she was having an irresistible urge to move her legs to relieve the sensations. The patient's symptoms repeated all day, and lasted for not less than 20 minutes, and if the patient did not stand up or change her position, it became exacerbated and intolerable. Symptoms worsened by night, and the severe nightly sleep disruption was significantly impairing her quality of life. The patient went to a gynecologist first because the vaginal symptoms disturbed the patient excessively. Her gynecologic evaluations were normal and she was referred to the neurology outpatient clinic for the problems in her legs. She had been a widow for 5 years. She had reached menopause four years previously. No symptoms of anxiety and depression were found in the clinical interview. Routine blood and urine tests, urine culture, and iron and ferritin levels were within normal limits. Her brain, pelvic and lumbosacral magnetic resonance imaging (MRI) were within normal limits. Electromyography excluded neuropathy of the pelvic nerves. The patient was started on pramipexole 0.25 mg/day, the symptoms diminished but did not resolve. The pramipexole dosage was gradually increased to 0.75 mg a day and all symptoms disappeared.

CASE 2

A 41-year-old female patient was admitted to the gynecology outpatient clinic with a feeling of arousal in the genital area for the last three months. The attacks lasted half an hour to 1 hour, and did not change day or night. The feeling of discomfort increased greatlywhen the patient crossed her legs. The patient constantly felt as if she needed to urinate, but her

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symptoms did not cease even if she emptied her bladder frequently. The patient thought she had a fungal infection because of the itching-like sensations. Although her physician found no signs of a fungal infection during the examination, he gave an antifungal vaginal suppository at the request of the patient. However, the patient had no relief. For this reason, she had been examined by three different gynecologists in the last 3 months. She was referred to a psychiatrist and a neurologist by her family doctor. Routine blood tests, ferritin levels, lumbosacral and pelvic MRI were performed. The patient had recently undergone a brain tomography for trauma, which was found as normal. She did not want to undergo pelvic electromyography. No additional pathology was detected except for a slightly low ferritin level. Low-dose pramipexole and ferrous sulphate was started. The dosage of pramipexole was increased to 0.75 mg/day because the patient reported no satisfactory improvement on 0.25 and 0.50 mg daily doses. Although the patient's symptoms were greatly reduced, the patient refused to use the pramipexole because of the nausea adverse effect. For this reason, she was started on pregabalin 75 mg per day and her symptoms completely resolved. At the routine follow-up at 6 months, it was learned that the patient had discontinued her medication for a while and her symptoms had recurred in the same way during this period. After restarting the pregabalin treatment, she was symptom free.

DISCUSSION

RGS is a very disturbing and embarrassing condition for patients. Coexistence with RLS and overactive bladder may be common. Although the pathophysiology of the disease is still unclear, neurologic, psychological or vascular theories have been proposed in the etiology. The possible underlying causes mentioned by Facelle et al. in their review were classified as psychological (associations with anxiety, depression, past sexual assault), biologic (pudendal nerve neuralgia, Tarlov cysts, pelvic varices), clinical clusters (RLS, overactive bladder), medication induced (selective serotonin reuptake inhibitors/serotonin–norepinephrine reuptake inhibitors, trazodone, nefazodone), and central nervous system (arteriovenous malformation, epilepsy, stroke) and dietary (soy intake) reasons (4).

Little is known about the frequency and causes of RGS, because all the data on this subject was collected from the literature, most of them from individual cases. Although there is no physical finding in many patients with RLS, case samples with morphologic changes in the etiology are not rare. Komisaruk and Lee collected sacral MRIs from an online support group for female PGAD, and Tarlov cysts were detected in 12 of 18 women who provided MR images (7). In a cohort of 1,045 patients with symptomatic spinal meningeal cysts, Feigenbaum et al. identified 8 of 11 women with PGAD symptoms had Tarlov cysts (8). According to Aquino et al. and Waldinger et al., this syndrome is in a cluster containing restless legs and / or overactive bladder syndrome (6, 9, 10). They believed that PGAD could be attributed to small-fiber neuropathy and that it could best be called 'restless genital syndrome.' After the examinations performed in our patients, we did not find any morphological cause in the etiology.

Our first patient was initially examined by a gynecologist. She came to our outpatient clinic because of the urge to move her legs after sitting or lying for a while. However, when the patient's symptoms were detailed, her genital symptoms were thought to be related to RLS. The second patient had been examined by 3 different gynecologists in the last 3 months and was finally referred to a psychiatrist and a neurologist by her family doctor.

As seen in previous studies, a dopaminergic mechanism seems to be important in the pathophysiology of the disease. Both of our patients were relieved with drugs used for RLS. Other treatment options based on the underlying causes may vary widely. These are cognitive behavioral treatments, psychotherapy, amitriptyline and other antidepressants, gabapentin, clonazepam, dopamine agonists, olanzapine, risperidone, anti-seizure medications (e.g. carbamazepine), varenicline (a high-affinity partial agonist for the $\alpha 4\beta 2$ nicotinic acetylcholine receptor subtype that leads to the release of dopamine in the nucleus accumbens), vascular venous embolization of pelvic varices, transcutaneous nerve stimulation, and pelvic floor massage (4, 11–13).

We want to draw attention to some notable points in the clinical diagnosis process of both patients. Neither patient thought that their symptoms might be of neurologic origin. Both patients reported that they wanted to be admitted by a female neurologist. It should be kept in mind that it may be very difficult to report a genital problem, especially in conservative societies.

In addition, when we examine the literature and question similar cases, we see that these patients do not know to which clinic branch they should present. Moreover, when they are examinedby a physician and tell them about their symptoms, they are not directed to neurology unless the symptoms belong to this branch. The presenting symptom is mostly referred to gynecology or psychiatry clinics and is not well recognized. In other words, although this disease has been described and entered the literature about 20 years ago, it is not sufficiently recognized either in our branch or among other clinical branches. In addition, there is no clear consensus in the treatment. It seems difficult to establish a clear consensus in this disease, which has only been described in very few cases and has many different etiologies.

Although the incidence of RGS is not known, it may be thought to be rare in clinical practice because patients do not think that the symptoms are neurologic or hesitate to report it because of embarrassment. This situation should be taken into consideration especially by colleagues working in outpatient clinics of gynecology, urology, and psychiatry.

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