Restless genital syndrome in a male patient relieved by pramipaxol and gabapentin

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Abstract

A 45 year old male sought consultation in our neurology clinic with the complaint of persistent genital uncomfortable sensations and pain. After extensive investigations, there was no underlying urological or neurological disease demonstrable. He was diagnosed to have restless genital syndrome and was given pramipexole and gabapentin with significant improvement of his symptoms. Our patient suggests that pramipexol and gabapentin may be useful as treatment for restless genital syndrome.

INTRODUCTION

Recent medical literature has identified an interesting diagnostic entity, termed "restless genital syndrome" (ReGS).1 This syndrome is mainly characterized by tingling, itching like uncomfortable sensations in the genital area.² This condition is mostly seen in female patients, though some male ReGS patients has also been reported. The diagnosis of ReGS requires the following diagnostic criteria: (i) Involuntary genital and clitoral arousal that persists for an extended period of time (hours, days, months); (ii) The physical genital arousal does not go away following one or more orgasms; (iii) The genital arousal is unrelated to subjective feelings of sexual desire; (iv) Yhe persistent feelings of genital arousal feel intrusive and unwanted; (v) Distress associated with persistent genital arousal.³

As mentioned,, only few male ReGS or persistent genital arousal disorder patients have been reported in the literature.⁴⁻⁵ There is limited knowledge on the treatment options and prognosis of these patients. We report here an adult male with ReGS Who responded to pramipexole and gabapentin.

CASE REPORT

A 45 year old male sought consultation in our neurology clinic with complaints of arousal and pain in the genital area which was worse at night. He reported that his symptoms started two years ago, and became worse recently. He complained of restlessness, intrusive tingling sensations and sometimes pain in the genital area. Recently these symptoms occurred almost every evening and extended into the late night. The symptoms did not occure during the daytime. This arousal was not related to sexual desire and was not triggered by both sexual and non-sexual stimuli. The patient reported that he felt irritable and desperate with these symptoms, and he used cold water in order to relieve the symptoms. He also added that standing or walking ameliorated his complaints. On the other hand, sitting and alcohol consumption aggravated the symptoms. He indicated that there was no problem in erection or ejaculation. Achieving orgasm did not relieve the genital sensations, but only resulted in temporary and partial relief. There was no significant past medical history except scrotal trauma 5 years earlier while playing amateur soccer. The patient also described restless leg syndrome symptoms in his mother. He did not smoke but consumed alcohol socially. The patient had no psychotic or depressive symptom. NSAID prescribed by an urologist did not relieve, and sertraline prescribed by the family physician aggravated the symtptoms.

The neurological and urological examination was normal. The blood tests including complete blood count, ferritin, urea, sugar, electrolytes, B12 vitamin, thyroid and hormonal screen were also normal. Electroencephalographic and electromyography examinations, cranial and pelvic magnetic resonance imaging, sonography of the urological system, and the urodynamic tests also showed no abnormality.

He was diagnosed as having ReGS, and was given pramipexole 0.25mg nocte. The dose was increased to 0.5 mg nocte after one week. There was significant decrease in the symptoms. Gabapentin 600mg nocte was then added to

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relieve the hyperesthesia, dysesthesia and improve sleep. All his intrusive and unwanted symptoms disappeared with the treatment.

DISCUSSION

The clinical features of our case were consistent with the diagnostic criteria of ReGS³, with physical arousal symptoms which were not related to sexual drive. The symptoms were ameliorated by pramipexole and gabapentin.

ReGS, earlier known as persistent sexual arousal syndrome, was first reported in medical literature by Leiblum and Nathan in 2001.6 In 2006, it was renamed as persistent genital arousal disorder by Goldmeierand Leiblum.⁷The prevalence, etiology, and pathogenesis of the syndrome are unknown.8 Nevertheless, many theories and etiological factors have been suggested. Amongst them are the central and peripheral neurological anomalies (including small fiber sensory neuropathy), vascular abnormalities causing pelvic congestion, mechanical impingement of the pudendal nerve, use or withdrawal of medications such as antidepressants, and psychological factors such as anxiety.9 Clitoral congestion and priapism was also suggested as a possible underlying cause for ReGS.¹⁰ The observations that sleep exacerbate the symptoms in ReGS may be explained by physiological increase in genital and pelvic congestion during sleep in the recumbent position. Sitting aggravated ReGS in 87% of patients, which may be due to a combination of pelvic congestion and/or neural compression.¹¹ Sitting also had the same effect in our patient.

There was report of male patient who responded to TENS application. This suggested peripheral nerve involvement in the etiology.⁵ Our patient responed to gabapentin, which is known to be effective against neuropathic pain. This also support the peripheral nerve component in the etiology of ReGS. Alcohol is a peripheral vasodilator. Aggravaton of symptoms with alcohol intake in our patient give support to the genital congestion hypothesis.¹²

There was no underlying structural etiology elicited in our patient after extensive urological and neurological examinations. There was also no psychiatric pathology demonstable to account for his symptoms. The aggravating effect of sertraline in our patient has already been noted previously.¹³ Relief of the symptoms with pramipexole and gabapentin suggests that it may be useful armetarian in the treatment of ReGS. In conclusion, we report here a male patient with ReGS whose symptoms were relieved by pramipexole and gabapentin.

DISCLOSURE

Conflict of interest: None

REFERENCES

- 1. Thubert T, Brondel M, Jousse M, *et al.* Persistent genital arousal disorder: a systematic review. *Prog Urol* 2012; 22(17):1043-50.
- Katsi V, Katsimichas T, Kallistratos MS, *et al.* The association of Restless Legs Syndrome with hypertension and cardiovascular disease. *Med Sci Monit* 2014; 20:654-9.
- Basson R, Leiblum S, Brotto L, et al. Definitions of women's sexual dysfunctions reconsidered: Advocating expansion and revision. J Psychosom Obstet Gynaecol 2003; 24(4):221-9.
- 4. Kamatchi R, Smith AA. Persistent genital arousal disorder in a male: a case report and analysis of the cause. *BJMP* 2013; 6:a605.
- Waldinger MD, Venema PL, van Gils AP, et al. Stronger evidence for small fiber sensory neuropathy in restless genital syndrome: two case reports in males. J Sex Med 2011; 8(1):325-30.
- Leiblum SR, Nathan SG. Persistent sexual arousal syndrome: A newly discovered pattern of female sexuality. J Sex Marital Ther 2001; 27(4):365-80.
- Goldmeier D, Leiblum SR. Persistent genital arousal in women—A new syndrome entity. *Int J STD AIDS* 2006; 17(4):215-6.
- 8. Waldinger MD, Schweitzer DH. Persistent genital arousal disorder in 18 Dutch women: Part II. A syndrome clustered with restless legs and overactive bladder. *J Sex Med* 2009; 6(2):482-97.
- Pink L, Rancourt V, Gordon A. Persistent genital arousal in women with pelvic and genital pain. J Obstet Gynaecol Can 2014; 36(4):324-30.
- Battaglia C and Venturoli S. Persistent genital arousal disorder and trazodone. Morphometric and vascular modifications of the clitoris. A case report. *Sex Med* 2009; 6(10): 2896-900.
- 11. Waldinger MD, Venema PL, van Gils AP, *et al.* New insights into restless genital syndrome: static mechanical hyperesthesia and neuropathy of the nervus dorsalis clitoridis. *J Sex Med* 2009; 6(10):2778-87.
- 12. Markos AR, Dinsmore W. Persistent genital arousal and restless genitalia: sexual dysfunction or subtype of vulvodynia? *Int J STD AIDS* 2013; 24(11):852-8.
- Rottach KG, Schaner BM, Kirch MH, *et al*.Restless legs syndrome as side effect of second generation antidepressants. *J Psychiatr Res* 2008; 43(1):70-5.