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New Insights into Restless Genital Syndrome: Static Mechanical Hyperesthesia and Neuropathy of the Nervus Dorsalis Clitoridis

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ABSTRACT ____

Introduction. Systematic study of dysesthetic and paresthetic regions contributing to persistent genital arousal in women with restless genital syndrome (ReGS) is needed for its clinical management.

Aim. To investigate distinct localizations of ReGS.

Methods. Twenty-three women, fulfilling all five criteria of persistent genital arousal disorder were included into the study. In-depth interviews, routine and hormonal investigations, electroencephalographs, and magnetic resonance imaging (MRI) of brain and pelvis were performed in all women. The localizations of genital sensations were investigated by physical examination of the ramus inferior of the public bone (RIPB) and by sensory testing of the skin of the genital area with a cotton swab (genital tactile mapping test or GTM test).

Main Outcome Measures. Sensitivity of RIPB, GTM test.

Results. Of 23 women included in the study, 18(78%), 16(69%), and 12(52%) reported restless legs syndrome, overactive bladder syndrome, and urethra hypersensitivity. Intolerance of tight clothes and underwear (allodynia or hyperpathia) was reported by 19 (83%) women. All women were diagnosed with ReGS. Sitting aggravated ReGS in 20(87%) women. In all women, MRI showed pelvic varices of different degree in the vagina (91%), labia minora and/or majora (35%), and uterus (30%). Finger touch investigation of the dorsal nerve of the clitoris (DNC) along the RIPB provoked ReGS in all women. Sensory testing showed unilateral and bilateral static mechanical Hyper-esthesia on various trigger points in the dermatome of the pudendal nerve, particularly in the part innervated by DNC, including pelvic bone. In three women, sensory testing induced an uninhibited orgasm during physical examination.

Conclusions. ReGS is highly associated with pelvic varices and with sensory neuropathy of the pudendal nerve and DNC, whose symptoms are suggestive for small fiber neuropathy (SFN). Physical examination for static mechanical Hyperesthesia is a diagnostic test for ReGS and is recommended for all individuals with complaints of persistent restless genital arousal in absence of sexual desire. Waldinger MD, Venema PL, van Gils APG, and Schweitzer DH. New insights into restless genital syndrome: Static mechanical hyperesthesia and neuropathy of the nervus dorsalis clitoridis. J Sex Med 2009;6:2778–2787.

Key Words. ReGS; RLS; OAB; Persistent Sexual Arousal Syndrome; PGAD; SFN; Pudendal Nerve; Dorsal Nerve of the Clitoris

Introduction

estless genital syndrome (ReGS) is character- \mathbf{K} ized by the five diagnostic criteria of persistent genital arousal disorder (PGAD) and the presence of restless legs syndrome (RLS) and/or overactive bladder (OAB) syndrome which occurrences are closely related to the genital symptoms [1,2]. The five diagnostic criteria of PGAD include: (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) the persistent feelings of genital arousal feel intrusive and unwanted; and (v) distress is associated with persistent genital arousal [3]. In 2001, persistent sexual arousal syndrome (PSAS) was for the first time reported in medical literature by Leiblum and Nathan [4]. In 2006, it has been renamed PGAD by Goldmeier and Leiblum [5]. The prevalence, etiology, and pathogenesis of the syndrome are unknown [1-5]. Current insight into PGAD has thus far been limited by a lack of systematic research, despite a number of case reports [4,6–16]. The first systematic medical study was recently conducted by Waldinger et al. [1,2] in 18 Dutch women fulfilling all five criteria of PGAD. It was found that the majority of women experienced PGAD during early menopause without preexisting psychiatric disorders and laboratory abnormalities. Most women had difficulties in describing the quality of the genital sensations, which were described in various terms and were diagnosed as dysesthesias and paresthesias. Electroencephalogram (EEG) analyses and brain magnetic resonance imaging (MRI) revealed no abnormalities, but MRI scans of the pelvis disclosed varices in 55% as confirmed by additional transvaginal ultrasonography [1,2]. The majority of women (67%) also reported preexistent or coexistent RLS, OAB, and hypersensitivity of the urethra. Notably, as a large burden of clinical evidence has pointed into the direction of a clinical cluster, this cluster of PGAD, RLS, OAB, and urethra hypersensitivity has been called restless genital syndrome [1,2]. In order to map the involved genital region in women with ReGS, we systematically examined this region, looking at tactile responses by means of a cotton swab. Each pressure point was evaluated by instructing women to report the evoked sensations. In the current study, we report on this test, which we have called genital tactile mapping test (GTM test).

Material and Methods

We prospectively evaluated 23 women with complaints of persistent unwanted feelings of genital arousal who visited our Outpatient Department of Neurosexology of HagaHospital Levenburg in The Hague between October 2004 and May 2009, and who were diagnosed with ReGS. The women were not actively recruited but were either referred by their general physician, gynecologist, and sexologist or contacted the first author after Internet information on the PGAD and ReGS research and treatment facilities in our Outpatient Department. According to the regulations of the medical ethical committee, official permission for study participation was not required as the study was not placebo controlled and study drugs were not taken. All patients were investigated by the first author, who followed an evaluation procedure according to standard protocol. After a neuropsychiatric and medical sexological interview of about 1 hour, women who were clinically diagnosed as having ReGS underwent routine and hormonal laboratory testing, an EEG, and an MRI scan examination of the brain and pelvis.

The diagnosis PGAD was established when the symptoms of the patients fulfilled all five criteria of PGAD [3]. Menopause was defined as the absence of menses for 12 months after the last menstruation. Perimenopause was defined as a combination of irregular cyclic bleedings and postmenopausal gonadotrophin levels. RLS was diagnosed according to the criteria of the International Restless Legs Syndrome Study Group [17]; (i) Urge to move the legs, usually accompanied or caused by uncomfortable leg sensations; (ii) Temporary relief with movement, partial or total relief from discomfort by walking or stretching; (iii) Onset or worsening of symptoms at rest or inactivity, such as when lying or sitting; and (iv) Worsening or onset of symptoms in the evening or at night. OAB was defined as increased urgency for micturition, with or without urge incontinence, with frequency and nocturia.

Between September 2008 and May 2009, the current study protocol of the GTM test was strictly followed by the first two authors in the presence of a nurse. This test for tactile sensations of the genital region is designed to analyze static mechanical pressure with a cotton swab at the skin near the vicinity of the genitals, perineum, anal area, groins, and pubic bone.

The effect of tactile stimuli with a cotton swab was systematically recorded. The test starts with

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