National Vulvodynia Association

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## Vulvodynia

**Botulinum Toxin A for Provoked Vestibulodynia: 12 Months' Follow-up of a Randomized Controlled Trial**

Philip Haraldson, Hanna Mühlrad, Ulrika Heddini, Kent Nilsson, Nina Bohm-Starke
J Sex Med 2022 Nov;19(11):1670-1679. doi: 10.1016/j.jsxm.2022.08.188. Epub 2022 Oct 26.
https://pubmed.ncbi.nlm.nih.gov/36307361/

**Background:** Provoked vestibulodynia (PVD) is a common pain disorder afflicting primarily young women, and botulinum toxin A (BTA) has been to a limited extent tested as a treatment. **Aim:** Evaluate outcome 12 months after injection with BTA as a treatment for PVD. **Methods:** We conducted a double-blinded, placebo-controlled trial of twice repeated injections of 50 units of BTA or placebo in the bulbocavernosus muscles, 3 months apart, in women with PVD. Treatment outcome after six months', failed to show any significant difference in pain reduction between the groups, as previously reported. Here, we report treatment outcomes 12 months after the first injections. In addition to injections, participants where instructed to perform pelvic floor exercises during month 6-12. 38 participants/group was calculated to achieve a statistical power of 80% based on an effect size of 20 VAS units (mean score range 56-76±31 SD). **Outcomes:** Primary outcome was self-reported dyspareunia or pain at tampon use, using a visual analogue scale (VAS) 0-100. Secondary outcomes were vaginal pressure measurements, psychological health, sexual function and distress. **Results:** From the initial 88 randomized women with PVD, 75 remained at 12 months; 38 in the BTA and 37 in the placebo group. There was no significant difference in primary outcome between the groups. Vaginal pressure in the BTA group had been restored to pre-treatment levels, with no differences between the groups at 12 months. There was an increase in sexual function in the BTA group, with a Female Sexual Function Index of 22.8 (±4.8) compared to the placebo group to 19.7 (±5.0), P=.048. No differences were observed in sexual distress, stress and anxiety. There was an increase in number of women attempting intercourse in the BTA group (74%) compared with placebo (43%), P=.005. Too few patients performed the pelvic floor exercises for this intervention to be analyzed. **Clinical implications:** This study highlights BTA as a safe treatment option for patients with PVD. **Strengths and limitations:** The randomized, double-blinded design and repeated treatments are the major strengths of this study and it is the first study to objectively evaluate muscular effect after BTA injections. The major shortcoming is that few participants performed the pelvic floor exercises, preventing analyses. **Conclusion:** At 12 months' follow up, no significant difference

in reduction of dyspareunia or pain at tampon use was observed. Women receiving BTA attempted intercourse more often and improved their sexual function compared with women receiving placebo.

**Vestibular hyaluronic acid injection in provoked vestibulodynia patients and its effect on pain and sexual function: A preliminary report**
Süleyman Eserdag, Burcu Akdag Özkok, Suat Süphan Ersahin, Emine Zeynep Yilmaz
Eur J Obstet Gynecol Reprod Biol. 2022 Nov 3;280:64-67. doi: 10.1016/j.ejogrb.2022.10.027.
https://pubmed.ncbi.nlm.nih.gov/36410243/

**Objective:** Provoked vestibulodynia (PVD) is a challenging and distressing problem for women. The aim of this study was to examine the effect of hyaluronic acid (HA) in the management of this condition. **Method:** This is a retrospective review of 12 women diagnosed with PVD and treated with HA (19 mg/mL) applied, point-by-point, to the vestibular region at 2 mm intervals and at a depth of 0.5 mm. Women completed a pain VAS and a Female Sexual Function Index (FSFI) before and 45 days after treatment. **Results:** An improvement was observed both in mean FSFI scores (17.8 to 23.3; p = 0.003) and mean VAS scores (7.2 to 4.1; p = 0.002) after HA application respectively. However, on a telephone interview 3 months post treatment, five women (41.7 %) complained of recurrence of their dyspareunia. **Conclusion:** HA is a promising management option in provoked vestibulodynia. However, further larger studies with possible alternative regimens and longer follow-up are required.

**Innervation of the human vulvar vestibule: A comprehensive review**
Leah Velikonja, Olivia Giovannetti, Michael A Adams, Diane Tomalty
Clin Anat. 2022 Oct 10. doi: 10.1002/ca.23966.
https://pubmed.ncbi.nlm.nih.gov/36216779/

Pain of the vulvar vestibule, including provoked vestibulodynia, is prevalent among women, yet challenging to treat due to its multifactorial etiology. Recent evidence indicates a neuroproliferative subtype in which hypersensitivity of the vulvar vestibule is due, in part, to hyperinnervation. Detailed knowledge regarding the innervation of the vulvar vestibule is crucial to understanding and treating pain conditions impacting this region. The purpose of this review is to consolidate the current evidence regarding the innervation of the human vulvar vestibule and discuss the implications of this innervation for pathological conditions affecting this tissue. A comprehensive review of the literature was conducted using keywords including vulvar vestibule, innervation, and vestibulodynia to identify articles concerning the innervation of the vulvar vestibule. Fifteen studies published between 1998 and 2017 were reviewed. Evidence from immunohistochemical investigations support that the vulvar vestibule has nociceptive, mechanosensory, sympathetic, and parasympathetic innervation. In pathological samples, hyperinnervation supports the neuroproliferative etiology of provoked vestibulodynia. Additionally, there is some evidence supporting the role of the pudendal nerve in vulvar vestibule innervation, although no cadaveric studies have been reported to date. Progress has been made in our understanding of the innervation of the vulvar vestibule, though further research into the origin of sensory and autonomic innervation of this region is needed. Advancing the knowledge of vulvar vestibule innervation is crucial towards improving our understanding of the function of this tissue, in addition to informing the etiology and management of pain syndromes impacting this region.

**A longitudinal case-control analysis of pain symptoms, fear of childbirth, and psychological well-being during pregnancy and postpartum among individuals with vulvodynia**
Kelly B Smith, Bozena Zdaniuk, Smruthi O Ramachandran, Lori A Brotto
Midwifery. 2022 Nov;114:103467. doi: 10.1016/j.midw.2022.103467. Epub 2022 Aug 23.
https://pubmed.ncbi.nlm.nih.gov/36108487/

**Objective:** Little research has examined changes in chronic vulvar pain (vulvodynia) symptoms with pregnancy and childbirth, nor fear as it relates to pregnancy/delivery amongst individuals with vulvodynia. The purpose of this study was to examine change in pain symptoms from pregnancy to postpartum amongst women with vulvodynia, as well as pain anxiety, fear of childbirth, and anxiety and depressive symptoms. **Design:** Prospective Case-Control Study. **Setting:** Online survey. **Participants:** Fifty-Seven pregnant individuals with a diagnosis of vulvodynia, and 41 pregnant control participants who reported being free of vulvar pain. Participants were recruited from the community and from hospital-based clinics for this study. **Measurements and findings:** Online surveys were administered to women diagnosed with vulvodynia and pain-free control participants during pregnancy and at three and six months postpartum. The survey contained both investigator-developed items and validated questionnaires, including the Pain Anxiety Symptoms Scale (PASS-20), the Wijma Delivery Expectancy/Experience Questionnaire (W-DEQ) to assess fear of childbirth, the Generalized Anxiety Disorder-7 (GAD-7) measure to assess symptoms of anxiety, and the Patient Health Questionnaire (PHQ-9) to assess symptoms of depression. Linear mixed models with random intercepts for longitudinal analyses indicated statistical improvements for most of the vulvar pain outcomes in the postpartum period amongst women with vulvodynia, including reduced pain intensity at three ($p = 0.005$) and six months ($p = 0.013$) postpartum for those women who delivered vaginally. The mean change in pain intensity corresponded though to only a minimal clinical change. Compared to controls, women with vulvodynia reported higher levels of fear of childbirth on the W-DEQ ($p = 0.024$). In both groups, increases in general anxiety on the GAD-7 were found from pregnancy to three ($p = 0.005$) and six months ($p = 0.033$) postpartum. Mode of birth moderated the findings for pain-related anxiety as measured by the PASS-20: only individuals who delivered via caesarean section reported increases in pain anxiety between pregnancy and six months postpartum ($p < 0.001$). **Key conclusions:** Pregnant women with vulvodynia experienced postpartum improvements in vulvar pain symptoms. Mode of birth may play a role in symptom trajectory. **Implications for practice:** Individuals with vulvodynia often have concerns about how pregnancy and childbirth will impact their symptoms. The current findings can be used to help such individuals make reproductive decisions knowing there may be improvements in vulvar pain and increases in anxiety that can occur postpartum. The statistical versus clinical significance of the pain intensity results also highlight the importance of asking each individual what changes in pain symptoms they experience and the meaning of such changes for that person.

**A multidisciplinary approach to a patient with vulvodynia: a successful treatment and outcome**
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Acta Dermatovenerol Alp Pannonica Adriat. 2022 Sep;31(3):119-121.
https://pubmed.ncbi.nlm.nih.gov/36149042/

Vulvodynia is chronic vulvar pain or a burning sensation lasting for at least 3 months without a cause. We present the case of a 53-year-old postmenopausal woman that experienced vulvar and vaginal burning, and discomfort and pain during sexual intercourse for 3 years, which greatly reduced her quality of life (QOL) despite the absence of itch and genital skin lesions. Her regular gynecological exams

showed no pathology, and so she was referred to a dermatologist, who initiated a multidisciplinary treatment approach involving several specialists: an anesthesiologist, gynecologist, urologist, psychiatrist, and dermatologist. Targeted psychiatric treatment (amitriptyline), together with acupuncture treatments and support by a gynecologist, led to a major improvement in symptoms and QOL, as well as a decrease in depression and anxiety measured by the Beck Depression Inventory II (BDI-II) and State-Trait Anxiety Inventory (STAI). A multidisciplinary and integrative approach was crucial for determining a diagnosis and achieving an excellent outcome.

## Chronic Pelvic Pain

**Pelvic Floor Physical Therapy and Its Merit in the Treatment of Female Urogenital Pain**
Annique Tracey
Curr Pain Headache Rep. 2022 Oct;26(10):775-782. doi: 10.1007/s11916-022-01076-0. Epub 2022 Sep 16.
https://pubmed.ncbi.nlm.nih.gov/36112273/

**Purpose of review:** Female urogenital pain (FUGP) affects many women and is often a diagnosis of exclusion. The long path to a diagnosis and subsequent treatment frequently leads to suffering on the individual's behalf (Obstet. Gynecol. 121: 645-50, 2013). Additionally, this delay in diagnosis and thus treatment places stress on the US medical system (Obstet. Gynecol. 121: 645-50, 2013). There is a lack of knowledge regarding the scope of pelvic floor physical therapy (PFPT) across the medical community that may prevent physicians from referring patients (J Urol. 193:1545-53, 2015; Sex Med Rev., 2021). PFPT is a low-risk, potentially high-reward option that should be recognized as part of the multidisciplinary approach to managing FUGP. **Recent findings:** Research databases (PubMed and Cochrane) were used to find articles on FUGP between 2005 and 2022. Systematic reviews, randomized controlled trials (RCTs), prospective and retrospective cohorts, and case-study analyses were included in reviewing the literature. The most recent studies in the last 2 years show the benefit of PFPT in certain FUGP diagnoses with improved pain scores and function when compared to no intervention or placebo treatment. The aim of this article is to elucidate the scope of PFPT in the treatment of FUGP with supporting research findings regarding efficacy. It is clear from the literature that PFPT should be recognized by referring physicians as part of a multidisciplinary approach to the treatment of FUGP.

**Retroperitoneal Causes of Genitourinary Pain Syndromes: Systematic Approach to Evaluation and Management**
Tarek Khalife, Amy M Hagen, Jessica E C Alm
Sex Med Rev. 2022 Oct;10(4):529-542. doi: 10.1016/j.sxmr.2022.06.009. Epub 2022 Sep 8.
https://pubmed.ncbi.nlm.nih.gov/36088274/

**Introduction:** Women with pelvic pain commonly report pain in their ovaries, vagina, uterus, or bladder. These symptoms may be caused by visceral genitourinary pain syndromes but also may be caused by musculoskeletal disorders of the abdomen and pelvis. Understanding neuroanatomical and musculoskeletal factors that may contribute to genitourinary pain is important for evaluation and management. **Objectives:** This review aims to (i) highlight the importance of clinical knowledge of pelvic neuroanatomy and sensory dermatomal distribution of the lower abdomen, pelvis, and lower extremities, exemplified in a clinical case; (ii) review common neuropathic and musculoskeletal causes

of acute and chronic pelvic pain that may be challenging to diagnose and manage; and (iii) discuss female genitourinary pain syndromes with a focus on retroperitoneal causes and treatment options. **Methods:** A comprehensive review of the literature was performed by searching the PubMed, Ovid Embase, MEDLINE, and Scopus databases using the keywords "chronic pelvic pain," "neuropathy," "neuropathic pain," "retroperitoneal schwannoma," "pudendal neuralgia," and "entrapment syndromes." **Results:** Retroperitoneal causes of genitourinary pain syndromes have substantial overlap with common conditions treated in a primary care setting. Thus, a comprehensive and systematic history and physical examination, with focused attention to the pelvic neuroanatomy, is key to establishing the correct diagnosis. In the clinical case, such a comprehensive approach led to the unexpected finding of a large retroperitoneal schwannoma. This case highlights the intricacy of pelvic pain syndromes and the complex nature of their possible overlapping causes, which ultimately affects treatment planning. **Conclusion:** Knowledge of the neuroanatomy and neurodermatomes of the abdomen and pelvis, in addition to understanding pain pathophysiology, is critical when evaluating patients with pelvic pain. Failure to apply proper evaluation and implement proper multidisciplinary management strategies contributes to unnecessary patient distress, decreased quality of life, and increased use of health care services. Khalife T, Hagen AM, Alm JEC. Retroperitoneal Causes of Genitourinary Pain Syndromes: Systematic Approach to Evaluation and Management.

**Novel anatomical findings with implications on the etiology of the piriformis syndrome**
Alexey Larionov, Peter Yotovski, Luis Filgueira
Surg Radiol Anat. 2022 Oct;44(10):1397-1407. doi: 10.1007/s00276-022-03023-5. Epub 2022 Sep 29.
https://pubmed.ncbi.nlm.nih.gov/36173479/

**Purpose:** The cause of the piriformis-related pelvic and extra-pelvic pain syndromes is still not well understood. Usually, the piriformis syndrome is seen as extra-pelvic sciatica caused by the entrapment of the sciatic nerve by the piriformis in its crossing through the greater sciatic foramen. However, the piriformis muscle may compress additional nerve structures in other regions and cause idiotypic pelvic pain, pelvic visceral pain, pudendal neuralgia, and pelvic organ dysfunction. There is still a lack of detailed description of the muscle origin, topography, and its possible relationships with the anterior branches of the sacral spinal nerves and with the sacral plexus. In this research, we aimed to characterize the topographic relationship of the piriformis with its surrounding anatomical structures, especially the anterior branches of the sacral spinal nerves and the sacral plexus in the pelvic cavity, as well as to estimate the possible role of anatomical piriformis variants in pelvic pain and extra-pelvic sciatica. **Methods:** Human cadaveric material was used accordingly to the Swiss Academy of Medical Science Guidelines adapted in 2021 and the Federal Act on Research involving Human Beings (Human Research ACT, HRA, status as 26, May 2021). All body donors gave written consent for using their bodies for teaching and research. 14 males and 26 females were included in this study. The age range varied from 64 to 97 years (mean 84 ± 10.7 years, median 88). **Results:** three variants of the sacral origin of the piriformis were found when referring to the relationship between the muscle and the anterior sacral foramen. Firstly, the medial muscle origin pattern and its complete covering of the anterior sacral foramen by the piriformis muscle is the most frequent anatomical variation (43% in males, 70% in females), probably with the most relevant clinical impact. This pattern may result in the compression of the anterior branches of the sacral spinal nerves when crossing the muscle.
**Conclusions:** These new anatomical findings may provide a better understanding of the complex piriformis and pelvic pain syndromes due to compression of the sacral spinal nerves with their somatic or autonomous (parasympathetic) qualities when crossing the piriformis.

## Persistent Genital Arousal Disorder

**Exploring the link between eating disorders and persistent genital arousal disorder/genito-pelvic dysesthesia: first description and a systematic review of the literature**
Hartmut Imgart, Annika Zanko, Sandra Lorek, Patti-Sue Schlichterle, Michael Zeiler
J Eat Disord. 2022 Nov 10;10(1):159. doi: 10.1186/s40337-022-00687-7.
https://pubmed.ncbi.nlm.nih.gov/36357896/

**Background:** Persistent Genital Arousal Disorder/Genito-Pelvic Dysesthesia (PGAD/GPD) characterized by recurrent physiological genital without corresponding psychological arousal is a poorly understood and researched condition. Based on the first two case descriptions of eating disorders directly linked to PGAD/GPD the aim of this paper was to systematically review the literature on possible associations between eating disorders and PGAD/GPD. **Method:** A systematic literature search on eating disorders and PGAD/GPD was conducted in PubMed, PsycINFO, and Scopus, complemented by Google Scholar. We included case reports, case series, cross-sectional studies and review articles published in peer-reviewed journals written in English or German-language. **Results:** The included original papers described a total of 2078 cases with PGAD/GPD symptomatology. Of these, 892 participants fulfilled all five PGAD/GPD core criteria. The aetiology of PGAD/GPD is unknown. Multifactorial genesis of PGAD/GPD is presumed including neurological, pharmacological, hormonal, vascular and psychological causes. A high degree of psychological comorbidity is reported. No study was found that drew a direct link between eating disorders and PGAD/GPD. Although PGAD/GPD symptoms also occur in adolescents, there are no findings in this regard. However, we found a gap in data collection: eating disorders as potential psychiatric comorbidities were systematically recorded in only a few studies. **Conclusion:** The existing literature have not yet considered a possible link between eating disorders and PGAD/GPD so far. According to the authors' knowledge, this work is the first review to systematically explore the associations. We suspect underreporting of PGAD/GPD cases in eating disorders and particularly during adolescence. We argue that there are several common factors that appear to be important in the etiology, course, and treatment of both disorders (e.g. hormonal dysregulation or sensory sensitivity and avoidance), warranting future research on the possible comorbidity of these disorders.

**Orgasm and Related Disorders Depend on Neural Inhibition Combined With Neural Excitation**
Barry R Komisaruk [1], Maria Cruz Rodriguez Del Cerro [2]
Sex Med Rev. 2022 Oct;10(4):481-492. doi: 10.1016/j.sxmr.2022.07.001.
https://pubmed.ncbi.nlm.nih.gov/36210092/

**Introduction:** Prevalent models of sexual desire, arousal and orgasm postulate that they result from an excitatory process, whereas disorders of sexual desire, arousal and orgasm result from an inhibitory process based on psychosocial, pharmacological, medical, and other factors. But neuronal excitation and active neuronal inhibition normally interact at variable intensities, concurrently and continuously. We propose herein that in conjunction with neuronal excitation, neuronal inhibition enables the generation of the intense, non-aversive pleasure of orgasm. When this interaction breaks down, pathology can result, as in disorders of sexual desire, arousal, and orgasm, and in anhedonia and pain. For perspective, we review some fundamental behavioral and (neuro-) physiological functions of neuronal excitation and inhibition in normal and pathological processes. **Objectives:** To review evidence that the variable balance between neuronal excitation and active neuronal inhibition at different intensities can account for orgasm and its disorders. **Methods:** We selected studies from searches on PubMed, Google Scholar,

Dialnet, and SciELO for terms including orgasm, neuronal development, Wallerian degeneration, prenatal stress, parental behavior, sensorimotor, neuronal excitation, neuronal inhibition, sensory deprivation, anhedonia, orgasmic disorder, hypoactive sexual desire disorder, persistent genital arousal disorder, sexual pain. **Results:** We provide evidence that the intensity of neuronal inhibition dynamically covaries concurrently with the intensity of neuronal excitation. Differences in these relative intensities can facilitate the understanding of orgasm and disorders of orgasm. **Conclusion:** Neuronal excitation and neuronal inhibition are normal, continuously active processes of the nervous system that are necessary for survival of neurons and the organism. The ability of genital sensory stimulation to induce concurrent neuronal inhibition enables the stimulation to attain the pleasurable, non-aversive, high intensity of excitation characteristic of orgasm. Excessive or deficient levels of neuronal inhibition relative to neuronal excitation may account for disorders of sexual desire, arousal and orgasm. Komisaruk BR, Rodriguez del Cerro MC. Orgasm and Related Disorders Depend on Neural Inhibition Combined With Neural Excitation.

## Pudendal Neuralgia

**Surgical management of pudendal nerve entrapment after sacrospinous ligament fixation**
Eva V Vodegel, Kim W M van Delft, Charlotte H C Nuboer, Claudia R Kowalik, Jan-Paul W R Roovers
BJOG. 2022 Oct;129(11):1908-1915. doi: 10.1111/1471-0528.17145. Epub 2022 Mar 29.
https://pubmed.ncbi.nlm.nih.gov/35289051/

**Objective:** To analyse the efficacy of sacrospinous ligament (SSL) suture removal on the reduction of pain symptoms in the case of suspected pudendal nerve entrapment after sacrospinous ligament fixation (SSLF). **Design:** Retrospective cohort study. **Setting:** Tertiary referral centre, the Netherlands. **Population:** A cohort of 21 women having their SSLF sutures removed because of SSLF-related pain symptoms. **Methods:** Clinical record review. **Main outcome measures:** The primary outcome was reduction of pain after SSL suture removal. Secondary outcome measures were time interval between suture placement and suture removal, complete suture removal, adverse events and recurrence of pelvic organ prolapse (POP). **Results:** A total of 21 women underwent SSL suture removal for severe and/or persistent pain, which was confirmed on clinical examination: 95% of the women (20/21) reported pain reduction after suture removal, and 57% reported complete pain relief. The time interval between suture placement and suture removal was at a median of 414 days (range 8-1855 days). Sutures could be completely removed in 86% of cases (18/21). One woman had excessive blood loss (520 ml) without blood transfusion. At 6-8 weeks after surgery, 10% of the women (2/21) had renewed symptomatic POP, stage ≥ 2, for which additional POP surgery was indicated. **Conclusions:** When performed by an experienced clinician, SSL suture removal is feasible and efficacious, with low morbidity. In addition, the risk of recurrent POP in the short term appeared to be low. **Tweetable abstract:** The surgical removal of sacrospinous ligament sutures is safe and efficacious for pain relief, even remote from initial placement.

**Risk Factors and a Nomogram for Prediction of Refractory Pudendal Neuralgia: A Retrospective Multivariate Analysis Study**
Xiao-Chen Wang, Long Wang, Yang Li, Gui-Jun Lu, Guo-Li Zhao, Ze-Guo Feng
Pain Physician. 2022 Sep;25(6):E815-E822.
https://pubmed.ncbi.nlm.nih.gov/36122264/

**Background:** Pudendal neuralgia (PN) is one of the most common forms of genital pain. About 4% or higher of patients suffering from chronic pain. **Objectives:** The aim of this study was to evaluate the risk factors for prediction of refractory PN (RPN).**Study design:** A retrospective multivariate analysis study. **Setting:** This retrospective analysis included 112 patients with PN who received the pudendal nerve block treatment at the Pain Department of General Hospital of People's Liberation Army. **Methods:** Univariate and multivariable logistic regression analyses were used for covariates selection. A nomogram was developed to estimate nonresponse to the pudendal nerve block. **Results:** The median age of patients and duration of patients were 48.0 and 1.25 years, respectively. Among 112 patients, there were 64 good responders to the pudendal nerve block for neuropathic pain and 48 non-responders. Multivariate analysis of 112 patients with PN demonstrated high self-rating depression scale scores (> 32) (odds ratio [OR], 95% confidence interval [CI]: 0.11, 0.01-0.77), damage to more than 2 terminal branches (OR, 95% CI: 0.22, 0.07-0.71), sensory deficit at S2-S4 on the dermatome map (OR, 95% CI: 0.22, 0.05-0.90), and duration of pain (> 4 years) (OR, 95% CI: 0.10, 0.03-0.42) were significant prognostic factors for nonresponse to the pudendal nerve block. **Limitations:** There are information biases for retrospective analysis, thus making it more difficult to come up with definitive conclusions. Large-scale randomized clinical trials are warranted to evaluate the risk factors for prediction of RPN. **Conclusions:** A longer duration of pain was correlated with a worse prognosis of the neurological disease. Patients with depression were prone to nonresponse to the pudendal nerve block treatment. Pain involved in more than 2 terminal branches and small fibers, affected at S2-S4 dermatome map, were considered to poor prognosis.

**Diagnosis and treatment of pudendal and inferior cluneal nerve entrapment syndrome: a narrative review**

Katleen Jottard, Pierre Bonnet, Viviane Thill, Stephane Ploteau, Stefan de Wachter

Acta Chir Belg. 2022 Dec;122(6):379-389. doi: 10.1080/00015458.2022.2123138. Epub 2022 Sep 16.
https://pubmed.ncbi.nlm.nih.gov/36074049/

**Aim:** Pudendal and inferior cluneal nerve entrapment can cause a neuropathic pain syndrome in the sensitive areas innervated by these nerves. Diagnosis is challenging and patients often suffer several years before diagnosis is made. The purpose of the review was to inform healthcare workers about this disease and to provide a basis of anatomy and physiopathology, to inform about diagnostic tools and invasive or non-invasive treatment modalities and outcome. **Methods:** A description of pudendal and inferior cluneal nerve anatomy is given. Physiopathology for entrapment is explained. Diagnostic criteria are described, and all non-invasive and invasive treatment options are discussed. **Results:** The Nantes criteria offer a solid basis for diagnosing this rare condition. Treatment should be offered in a pluri-disciplinary setting and consists of avoidance of painful stimuli, physiotherapy, psychotherapy, pharmacological treatment led by tricyclic antidepressants and anticonvulsants. Nerve blocks are efficient at short term and serve mainly as a diagnostic tool. Pulsed radiofrequency (PRF) is described as a successful treatment option for pudendal neuralgia in patients non-responding to non-invasive treatment. If all other treatments fail, surgery can be offered. Different surgical procedures exist but only the open transgluteal approach has proven its efficacy compared to medical treatment. The minimal-invasive ENTRAMI technique offers the possibility to combine nerve release with pudendal neuromodulation. **Conclusions:** Pudendal and inferior cluneal nerve entrapment syndrome are a challenge not only for diagnosis but also for treatment. Different non-invasive and invasive treatment options exist and should be offered in a pluri-disciplinary setting.

**When and How to Utilize Pudendal Nerve Blocks for Treatment of Pudendal Neuralgia**
Abigail Cain, Kimberly Carter, Christina Salazar, Amy Young
Clin Obstet Gynecol. 2022 Dec 1;65(4):686-698. doi: 10.1097/GRF.0000000000000715. Epub 2022 Jun 3.
https://pubmed.ncbi.nlm.nih.gov/35703212/

Chronic pelvic pain is a common cause of pain in reproductive age women with debilitating consequences for affected women's health and quality of life. Treatment providers must be well versed in all treatment options for these patients, understanding the overlap in the management and treatment of chronic pelvic pain caused by pudendal neuralgia, myofascial pelvic pain, and vulvodynia. Pudendal blocks are a simple and quick procedure that can be performed in the office and often helps improve all the above conditions when used along with other treatment options. We review the anatomy and methodology on when and how to perform pudendal blocks in the office to better inform the general gynecologist on how to implement offering this treatment in the outpatient clinical setting.

**Unidentified Branches of the Posterior Femoral Cutaneous Nerve and Persistent Neuropathy**
Michelle R Jennette, David Bailey, Neel Patel, Elias Rizk
Cureus. 2022 Sep 22;14(9):e29447. doi: 10.7759/cureus.29447. eCollection 2022 Sep.
https://pubmed.ncbi.nlm.nih.gov/36299977/

The posterior femoral cutaneous nerve (PFCN) is an extensive nerve with numerous collateral branches which provide cutaneous innervation to 2/3$^{rds}$ of the posterior thigh, the infragluteal fold, as well as the lateral anal region, scrotum, and labia majora through its inferior cluneal and pudendal nerve branches. It has been noted in multiple studies that patients can experience persistent PFCN neuropathy after surgery for decompression of known collateral branches. In this study, we used 17 formaldehyde (7 male and 10 female) perfused cadavers obtained from Hershey Medical Center's donor program to study the branching patterns of the PFCN. As a result, we found that 41% of individuals have an unidentified proximal branch of PFCN that recurs over the inferolateral border of the gluteus maximus, suggesting other areas of potential compression or nerve entrapment that could lead to persistent PFCN neuropathy that's not improved after treatment for sciatic, pudendal, or inferior cluneal neuralgia. We hope these findings allow clinicians to modify current surgical techniques and improve patients' post-operative quality of life.

**Right laparoscopic pudendal release + neurostimulator prosthesis (LION procedure) in pudendal neuralgia**
Enrique Moncada, Alberto de San Ildefonso, Erene Flores, Lucia Garrido, Oscar Cano-Valderrama, Vincenzo Vigorita, Raquel Sánchez-Santos
Colorectal Dis. 2022 Oct;24(10):1243-1244. doi: 10.1111/codi.16190. Epub 2022 Jun 27.
https://pubmed.ncbi.nlm.nih.gov/35575432/

**Aim:** Pudendal neuralgia is a highly disabling entity with complex diagnostic and controversial treatment results. Surgical neurolysis has been shown to be the most effective treatment. Sacral root neurostimulation or posterior tibial nerve stimulation are used to rescue patients who either have not responded to surgery or have worsened after an initial improvement. **Methods:** Given the excellent visualization of the pudendal nerve during laparoscopic pudendal release, we propose to combine this procedure with neurostimulation, taking advantage of the possibility of in situ placement of the electrode. The abdominal cavity is accessed laparoscopically through four ports, and after identifying

and releasing the pudendal nerve a neurostimulation electrode is placed next to the nerve and is connected to a generator located in a subcutaneous pocket. **Results:** This procedure has been performed in one patient with a satisfactory result. **Conclusions:** Laparoscopic pudendal release with neurostimulator prosthesis is an experimental technique that can be promising for the treatment of pudendal neuralgia.

## Dermatological Conditions

**Clinical study on multi-focused laser in the treatment of vulvar lichen sclerosus**
Jing-Qiu Guo, Song-Yan Chen, Xue-Mei Chen, Jing-Quan Lu, Yu Song, He-Yu Liu, Li-Na Hu, Zheng-Yan Zhu
Front Surg. 2022 Sep 15;9:919135. doi: 10.3389/fsurg.2022.919135. eCollection 2022.
https://pubmed.ncbi.nlm.nih.gov/36189386/

**Objective:** To investigate the clinical effect of Multi-focused (MF) laser in the treatment of vulvar lichen sclerosus (VLS). **Methods:** In this single-center, randomized controlled trial, we compared the effect of fractionated MF laser with other treatments on patients with biopsy-proven VLS. Patients with VLS were enrolled in this study and randomly divided into three groups. Patients in the experimental group were treated with a $CO_2$ laser, control group 1 was treated with radiofrequency, and control group 2 was treated topically with glucocorticoids and soaking with Chinese patent medicine. The pruritus degree, skin elasticity, skin color, lesion scope, and total score were compared before treatment, at one month after treatment, and three months after treatment. **Results:** One month after treatment, the pruritus degree, skin elasticity, skin color, lesion scope, and total score decreased in the experimental group, and the differences were statistically significant ($P < 0.05$). In control group 1, the differences in pruritus degree, skin color, and total score were statistically significant ($P < 0.05$), but the differences in skin elasticity and lesion scope were not statistically significant ($P > 0.05$). In control group 2, the differences in pruritus degree and total score were statistically significant ($P < 0.05$), but the differences in skin elasticity, skin color, and lesion scope were not statistically significant ($P > 0.05$). At one month after the end of treatment, the differences in pruritus degree, skin elasticity, skin color, lesion scope, and total score among the three groups were not statistically significant. At three months after the end of treatment, the differences in the scores of the five indicators were statistically significant.
**Conclusion:** For the three treatment methods for VLS, topical corticosteroids + traditional Chinese medicine can quickly relieve itching symptoms in patients, but it cannot significantly improve skin elasticity, skin color, and lesion scope, and VLS easily relapses after treatment. Radiofrequency can improve itching symptoms and skin color but has poor effects on the change of skin elasticity and lesion scope. Multi-focused laser treatment can alleviate the degree of pruritus, improve skin color and elasticity, and narrow the lesion scope, and VLS will not relapse within three months after treatment.

**Vulvar dermatoses: a cross-sectional 5-year study. Experience in a specialized vulvar unit**
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**Background:** Vulvar diseases are common in the general population and have a negative impact on the quality of life. **Objectives:** To describe our experience as dermatologists in the management of vulvar dermatosis consultations. **Methods:** A retrospective observational study was conducted with patients who attended monographic vulvar consultations over a 5-year period. Clinical information was obtained from the patient's charts. **Results:** 148 women were studied. Their mean age was 43.24 years (standard deviation: 15.15 years), with ages ranging from 4 months to 80 years. 53.4% of patients took between 2 and 5 years to seek medical attention for the first time. The most frequent diagnosis was lichen sclerosus (41.9%), irritative eczema of the vulva (14.9%), and lichen simplex chronicus (10.1%). 83.8% reported anogenital itching, 66.2% pain, and 45.9% dyspareunia. The most frequently prescribed treatment was ultra-potent topical corticosteroids (clobetasol propionate; 41.2%). Patients with lichen sclerosus were significantly older than those who presented with any of the other diseases. No differences were found in terms of either the time of disease evolution or in symptom presentation. **Study limitations:** Retrospective study. Vulvar diseases with an infectious cause are usually managed in primary care, therefore, were not included. All patients were recruited from a single private hospital which limits the comparisons with the public health system. **Conclusions:** Vulvar diseases frequently occur and are associated with high morbidity. It is essential to promote the development of specific vulvar consultations in hospitals. Specialties such as dermatology, gynecology, urology, or physiotherapy must be part of these units.

**Does Clearance of Vulvar Lichen Sclerosus after a Corticosteroid Treatment Correspond to a Decrease in Disease-Related Burden? Results from a Cohort Study Using Pictorial Representation of Illness and Self-Measure and the Dermatology Life Quality Index**

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**Background and objectives:** Complete clearance of vulvar lichen sclerosus (VLS) occurs in a minority of treated patients. Disease persistence may impact patient well-being. The main objective of this study was to assess if achieving a complete clearance with a corticosteroid treatment leads to a benefit in terms of patient suffering and quality-of-life (QoL) impairment. **Methods:** We performed an observational study on a cohort of VLS women, who applied mometasone furoate 0.1% ointment for 12 weeks. At treatment completion (T1), we compared the patients who achieved clearance in symptoms (Global Subjective Score [GSS] = 0) or in objective features (Global Objective Score [GOS] = 0) or in both with those who achieved a lower degree of improvement, on the basis of Pictorial Representation of Illness and Self-Measure (PRISM) and Dermatology Life Quality Index (DLQI) scores.**Results:** In the whole sample (n = 101), GSS, GOS, PRISM, and DLQI scores significantly improved after treatment from baseline; 34 patients (35.8%) achieved GSS = 0, 26 (25.7%) achieved GOS = 0, and 11 (11.5%) clearance of GSS and GOS. PRISM scores at T1 were significantly higher in patients who achieved clearance of symptoms when compared with those who did not, including patients achieving 50-99% GSS improvement from baseline. DLQI scores were lower in patients who achieved clearance of symptoms, signs, or both when compared with the others. **Conclusions:** VLS clearance corresponded to a significant improvement in the QoL of VLS patients, also in comparison with those who achieved a substantial but incomplete decrease of symptom and sign scores, and should become an ideal therapeutic goal.

**Why do some patients with vulval lichen sclerosus on long-term topical corticosteroid treatment experience ongoing poor quality of life?**

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**Objective:** To identify reasons for ongoing poor quality of life (pQOL) in a subset of long-term topical corticosteroid-treated vulval lichen sclerosus (VLS) patients. **Methods:** A prospective cross-sectional study of patients attending a dermato-gynaecology practice in Sydney, Australia, comparing VLS patients with good quality of life (gQOL) and pQOL, in pre-treatment and long-term treatment groups, using the Vulval Quality of Life Index (VQLI). Demographics, VQLI scores and treatment characteristics were compared between gQOL and pQOL patients. **Results:** A total of 255 biopsy-proven VLS patients, 67 in pre-treatment and 188 in long-term treated groups were considered. There were 33 (49.3%) pQOL patients in pre-treatment and 13 (6.9%) in treatment groups ($p < 0.001$). The highest-scoring domain in treated pQOL patients was sexuality (1.7 [interquartile range (IQR) 1.0-2.0]), followed by anxiety [1.3 (IQR 1.0-1.5]), symptoms (1.0 [IQR 0.5-1.5]) and activities of daily living (0.7 [IQR 0.3-1.0]). Compared to treated gQOL, treated pQOL had significantly higher proportions of patients with partial treatment adherence (8 [61.5%] vs 42 [24.0%], $p = 0.006$), suboptimal disease control (7 [53.8%] vs 20 [11.4%], $p < 0.001$), scarring progression (3 [23.1%] vs 7 [4.0%], $p = 0.024$) and urinary incontinence (5 [38.5%] vs 27 [15.4%], $p = 0.049$). **Conclusions:** Only a minority of long-term treated VLS patients reported ongoing pQOL. Of those who did, sexuality and anxiety domains were found to be the main sources of distress. Three major areas distinguishing gQOL from pQOL patients were (1) treatment adherence and disease control, (2) psychological factors and (3) urinary incontinence.

**In vivo evaluation of vulvar lichen sclerosus with reflectance confocal microscopy and therapeutic monitoring in children**

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**Background:** Vulvar lichen sclerosus (VLS) in girls presents with itching, dysuria, and constipation and may result in the loss of vulvar architecture. In patients with an ambiguous clinical presentation, reflectance confocal microscopy (RCM) could be a helpful noninvasive diagnostic tool. The aim of this study was to describe the RCM characteristics of VLS and explore the clinical application value of RCM in therapeutic monitoring. **Methods:** Sixteen patients with VLS were included in the study. All patients were periodically evaluated clinically with RCM, and different treatment regimens were given based on the patient's clinical appearances and RCM features. **Results:** Some major key diagnostic features of VLS can be observed by RCM, including round to oval cyst-like structures with medium-to-low-refractive keratinoid substances (75%), thinning of the epidermal thickness (100%), destruction of the ring-like structures around dermal papillae (100%), disorderly distributed coarse medium-refractive fibrous material (100%),polygonal, plump, high-refractive cellular structures and linear low-refractive canalicular structures (100%). All of these characteristics had a high correspondence with histopathological features. The clinical manifestations improved after individualized treatment regimens based on the clinical appearances and RCM features. **Conclusion:** RCM allows the visualization of major key diagnostic features of VLS and represents a valid option for objective therapeutic monitoring.

**Characterization of patients with vulvar lichen sclerosus and association to vulvar carcinoma: a retrospective single center analysis**

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**Purpose:** Lichen sclerosus (LS) is a benign, cutaneous, chronic inflammatory (autoimmunological) disease. The differentiated vulvar intraepithelial neoplasia (dVIN) accounts for a precursor lesion of vulvar squamous cell carcinoma and is often associated with lichen sclerosus. Although the association between lichen sclerosus and vulvar carcinoma has long been recognized, there is a lack of evidence in literature. **Methods:** This retrospective study examined pseudonymized data of 499 women diagnosed with vulvar pathology between 2008 and 2020 at the Department of Gynaecology and Obstetrics of Hannover Medical School (MHH). Data were further stratified for the time of onset, location of disease, accompanying disease, HPV status and progression of disease into vulvar squamous cell carcinoma (VSCC). **Results:** In total, 56 patients were diagnosed with vulvar lichen sclerosus. The mean onset of disease was at 60.3 years of age. After subdividing cases of diagnosed LS into those who did not develop vulvar carcinoma in their course and those who did, the ages at onset are 52.66 ± 17.35 and 68.41 ± 10.87, respectively. The incidence of vulvar cancer in women diagnosed with lichen sclerosus was 48.2%. Twenty-five patients reported a diagnosis of VIN in their self-reported history. **Conclusions:** In our retrospective study, we showed a trend between vulvar lichen sclerosus and VSCC. The difference between the two age groups of patients diagnosed with lichen sclerosus who developed vulvar carcinoma and those who did not is statistically significant. Our results highlight the importance to diagnose lichen sclerosus early to ensure adequate follow-up and prevent progression to VSCC.

**Lichen sclerosus and immune checkpoint inhibitors: A case and review of the literature**

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Up to 50% of patients treated with immune checkpoint inhibitors (ICIs) develop cutaneous immune-related adverse events (cirAEs).[1] Genital lichen sclerosus (GLS) and extragenital lichen sclerosus (ELS) are rarely reported as cirAEs.[1] We report a woman who presented with both GLS and ELS while being treated with ICIs and performed a literature review of the association of both conditions with ICIs (methodology in Supplementary Material S1). Informed consent was obtained.

Our patient was a 54-year-old female who developed vulvar pruritus and an asymptomatic cutaneous lesion on her abdomen 7 months after commencing adjuvant treatment with nivolumab 240 mg Q2W plus ipilimumab 1 mg/kg Q6W for her stage 3B left arm melanoma. She experienced menopause at 51 and was otherwise well with no personal or family history of autoimmune conditions. On examination, there were clinical features of GLS and ELA (Figure **1a,b**). A biopsy of the abdomen showed histological features of lichen sclerosus (Figure **1c**). She was prescribed clobetasol propionate 0.05% ointment daily for a month with a decreasing regimen, which achieved symptom control. Her ELS was mild, and the patient elected no treatment. Four years after completing adjuvant immunotherapy, the patient remains in complete remission and is regularly reviewed for her GLS.