A Novel Collaborative Protocol for Successful Management of Penile Pain Mediated by Radiculitis of Sacral Spinal Nerve Roots From Tarlov Cysts

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ABSTRACT

Introduction: Since 14 years of age, the patient had experienced extreme penile pain within seconds of initial sexual arousal through masturbation. Penile pain was so severe that he rarely proceeded to orgasm or ejaculation. After 7 years of undergoing multiple unsuccessful treatments, he was concerned for his long-term mental health and for his future ability to have relationships.

Aim: To describe a novel collaboration among specialists in sexual medicine, neurophysiology, and spine surgery that led to successful management.

Methods: Collaborating health care providers conferred with the referring physician, patient, and parents and included a review of all medical records.

Main Outcome Measure: Elimination of postpubertal intense penile pain during sexual arousal.

Results: The patient presented to our sexual medicine facility at 21 years of age. The sexual medicine physician identifying the sexual health complaint noted a pelvic magnetic resonance imaging report of an incidental sacral Tarlov cyst. A subsequent sacral magnetic resonance image showed four sacral Tarlov cysts, with the largest measuring 18 mm. Neuro-genital testing result were abnormal. The neurophysiologist hypothesized the patient's pain at erection was produced by Tarlov cyst-induced neuropathic irritation of sensory fibers that course within the pelvic nerve. The spine surgeon directed a diagnostic injection of bupivacaine to the sacral nerve roots and subsequently morphine to the conus medullaris of the spinal cord. The bupivacaine produced general penile numbness; the morphine selectively decreased penile pain symptoms during sexual arousal without blocking penile skin sensation. The collaboration among specialties led to the conclusion that the Tarlov cysts were pathophysiologically mediating the penile pain symptoms during arousal. Long-term follow-up after surgical repair showed complete symptom elimination at 18 months after treatment.

Conclusion: This case provides evidence that (i) Tarlov cysts can cause sacral spinal nerve root radiculitis through sensory pelvic nerve and (ii) there are management benefits from collaboration among sexual medicine, neurophysiology, and spine surgery subspecialties. **Goldstein I, Komisaruk BR, Rubin RS, et al. A Novel Collaborative Protocol for Successful Management of Penile Pain Mediated by Radiculitis of Sacral Spinal Nerve Roots From Tarlov Cysts. Sex Med 2017;5:e203–e211.**

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Key Words: Tarlov Cyst; Neurogenic Sexual Dysfunction; Penile Pain; Radiculitis of Sacral Spinal Nerve Root

INTRODUCTION

Male genital organs, including the penis, corpora cavernosa, corpus spongiosum, pelvic floor muscles, prostate, and scrotum, are innervated in part by the afferent (sensory) and efferent (motor) components of the pudendal somatic, pelvic, and hypogastric autonomic nerves.[1–3]

Injuries and neuropathies of the somatic and/or autonomic genital nerves can occur in the following well-described and well-recognized conditions: post-radical prostatectomy, multiple sclerosis, diabetes mellitus, traumatic pudendal neuropathy, and spinal cord injury.[4–7] Such injuries and neuropathies to the

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critical somatic and autonomic genital nerves could hypothetically irritate and/or block genital nerve function, which, depending on their location, intensity, and chronicity, can result in different distressing sexual health concerns.

Injuries and neuropathies of the somatic and/or autonomic genital nerves also can occur from less well-described and less well-recognized conditions. For example, in the sacral and lumbar spine regions, sacral spinal nerve roots are subject to compression or impingement radiculopathies from pathologies such as sacral spinal meningeal cysts (eg, Tarlov cysts), disc impingements, annular tears, facet (spinal synovial) cysts, and spinal stenosis.[8–11]

Should these sacral spinal nerve roots become compressed and/or irritated, neurologic sexual dysfunctions secondary to radiculopathy of the sacral spinal nerve roots can be realized.[8] Of critical importance, and having a uniquely different prognosis from such conditions as post-radical prostatectomy or diabetes mellitus, sacral spinal nerve root compression injuries and neuropathies are potentially reversible by spine surgery.[12]

This case report highlights evidence that Tarlov cysts can pathophysiologically produce the extremely distressing sexual health concern of painful penile genital neuropathy induced by sexual arousal. Previously, Tarlov cysts have been linked to cases of persistent genital arousal disorder (PGAD) in women.[8,12] This case report also emphasizes the management benefits of a collaborative association among experts in three disciplines: sexual medicine, neurophysiology, and spine surgery. Such an association was used to successfully manage this young patient who presented to our sexual medicine clinic with a 7-year history of severe, distressing penile pain that commenced within seconds of starting masturbation.

CASE REPORT

The patient reported that the condition started when he was 14 years old, before his beginning masturbation, when he would awaken once or twice per month with painful erections without nocturnal emissions. He subsequently noted that within seconds of starting masturbation, he repeatedly experienced severe penile pain that was so intense that it prevented him on almost all occasions from proceeding to orgasm. He perceived the pain exclusively in the penile shaft, not in the glans penis or surrounding genitalia (ie, testes, scrotum, perineum, pelvic floor muscles, anus, or prostate). Voluntary pelvic floor contraction was not painful. Voiding and defecation did not elicit pain. The intense pain ceased within seconds after terminating erect penile stimulation. With the onset of infrequent nocturnal erections, the intense pain of his penis would wake him up. Although he almost never experienced a sexually induced orgasm, on those rare occasions when he did, he reported that even the passage of ejaculate fluid through the urethra was painful. He also experienced urinary urgency and complained of restless legs.

The patient experienced two possibly relevant etiologic episodes of trauma: when he fell down the stairs at 8 months of age and when he fell onto, straddling, the metal bar of the "monkey bars" at 7 years of age, injuring his perineal and coccygeal areas.

After presenting with symptoms at 14 years of age, the patient and his very supportive parents, strong advocates in communicating with the various physicians, sought medical assessments during the next 7 years. During his adolescent years, he was managed by specialists in chiropractic medicine, urology, neurology, pain medicine, physical medicine and rehabilitation, pelvic floor physical therapy, sexual medicine, psychiatry, and sex therapy with cognitive behavior therapy. Based on numerous careful physical examinations, ultrasound, and magnetic resonance imaging (MRI) studies, there was no evidence of tender Peyronie disease plaques; no apparent prostate, pelvic, scrotal, abdominal, or inguinal pathology; no tethered spinal cord or bladder or bowel pathology; and no evidence of a sexually transmitted infection; also normal were endocrine function, including testosterone, and non-hormonal blood test results. One pain specialist admitted to "no answers, no suggestions, and no tests that can be done." From multiple physicians over the years, the patient was prescribed the following medications, none of which alleviated the complaint of pain after initiation of penile erection by masturbation: pregabalin, lorazepam, paroxetine, nortriptyline, diazepam, doxazosin, acetaminophen, acetaminophen with codeine, a eutectic mixture of lidocaine 2.5% and prilocaine 2.5%, and botulinum toxin type A injected into the bulbospongiosus muscles. On separate occasions with different specialists, he received a dorsal nerve block and pudendal nerve blocks with local steroid triamcinolone peripherally in the perineum and at the Alcock canal. All these invasive procedures failed to block his symptom of intense penile pain at masturbation. In an additional procedure, lidocaine subcutaneously around the neurovascular bundle blocked not only the pain but also all penile sensibility, producing penile numbness and a lack of erection.

The penile pain with sexual stimulation discouraged him from initiating sexual relationships. He began to struggle emotionally at 19 years of age when he went to college. A sexual medicine psychiatrist assessed him and judged he did not present as overly anxious or histrionic in any way, but the clinician was concerned about emerging anxiety and depression as the young man came to the realization that this condition was not going to resolve naturally with time. The psychiatrist did not find sufficient psychiatric pathology to account for his pain. The patient's consultations with medical experts continued with the support of his parents.

At 21 years of age, after 7 years of examinations, he sought an opinion at a sexual medicine facility. His baseline sexual function questionnaire data were noted (Table 1). Included in the medical records was a pelvic MRI and a prostate MRI radiology report noting the presence of a Tarlov cyst on S2, listed as "incidental" (Figure 1). A sacral MRI, which can better detect spinal pathology not visible on a pelvic MRI, was obtained with T2

Table 1. Successful treatment results of patient-reported outcomes at baseline on entry to the sexual medicine clinic and then 18 months postoperatively

Questionnaire	Baseline	18 mo postoperatively	Outcome changes
IIEF erectile function domain (maximum score = 30)	18	30	mild to moderate dysfunction → no dysfunction
IIEF orgasm function domain (maximum score = 10)	2	10	severe dysfunction → no dysfunction
IIEF sexual desire domain (maximum score = 10)	9	9	no dysfunction
IIEF intercourse satisfaction domain (maximum score = 15)	0	11	was not having intercourse → no dysfunction
IIEF sexual satisfaction domain (maximum score = 10)	2	7	severe dysfunction → mild dysfunction
IIEF total score (maximum score = 75)	31	67	marked improvement of sexual function
Sexual Distress Scale—Revised (maximum score = 52)	36	8	marked decrease of sexual distress
PSS (maximum score = 40)	19	17	mild decrease in perceived stress levels
PHQ-9 (maximum score = 27)	7	3	mild depression → minimal depression
McGill Pain Questionnaire—Intensity (maximum score = 5)	5	0	pain = 5 of 5 → 0 of 5

IIEF = International Index of Erectile Function[24]; PHQ-9 = Perceived Health Questionnaire[27]; PSS = Perceived Stress Scale.[26]

weighting, sagittal, coronal, and axial views, with and without contrast. The sacral MRI showed four Tarlov cysts located at the right and left S2 and S3 dorsal roots: 18 mm on the right at S2, 15 mm on the left at S2, and 8 mm each on the right and left at S3 (Figure 2).

The patient underwent subsequent neuro-genital testing.[13–15] Quantitative sensory testing was performed on a non-genital control site (the pulp of the right index finger) and on multiple penile sites (glans penis, right lateral shaft, and left lateral shaft of penis) with biothesiometry for determination of vibration perception and with a thermal sensitivity testing instrument for determination of hot and cold thresholds (ie, "Say when it first feels hot or cold").[16,17] Compared with the non-genital control site, quantitative sensory testing disclosed increased vibratory, cold, and heat perception threshold values consistent with mild to moderate sensory neuropathy of the dorsal nerve branches of the pudendal nerve or afferent sacral root. Insertion of a lubricated urethral self-catheter into the midurethra tests the integrity of the pudendal and pelvic nerves.[18] The patient underwent this procedure including administration of sterile saline 5-10 mL into the urethra. He reported being very aware of the pain in addition to discomfort from urethral distension.

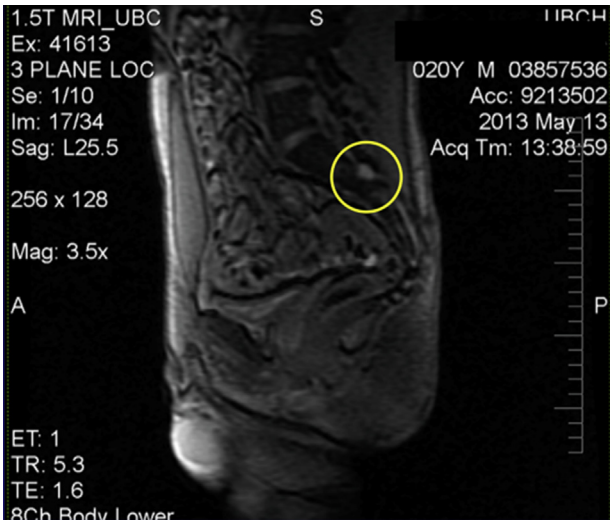

Figure 1. Pelvic magnetic resonance image shows a limited view of the Tarlov cyst (yellow circle). It is the sacral magnetic resonance image that specifically visualizes the anatomic orientation of the nerve roots in relation to the sacrum. The goal of sacral vs pelvic magnetic resonance imaging is to obtain sequences that identify the anatomic characteristics of the Tarlov cyst.

The neurophysiologist reported that previously a correlation had been shown between the presence of Tarlov cysts in women and the presence of PGAD.[8] He further emphasized that the pudendal nerves and pelvic nerves provide sensory afferent innervation from the penis. These afferent nerves enter the spine at the S2 to S4 levels.[19] A study by Komisaruk and Lee[8] raised the possibility that there could be a correlation between the Tarlov cyst and the patient's intense penile pain in relation to the initiation of sexual arousal during masturbation. The neurophysiologist suggested that, in this case, the Tarlov cysts could be the pathophysiologic basis for his penile pain symptoms, in particular stretching, abrading against the sacral foramina, and thereby irritating the sensory afferent component of the pelvic autonomic nerves transmitting penile distension.

Thus, the patient was advised to undergo a diagnostic test for the possible etiologic role of the Tarlov cysts. Using computed tomographic guidance and local anesthesia, the spine surgeon injected 2 mL of the local anesthetic, 0.05% bupivacaine, at the level of the S2 and S3 dorsal roots at their point of entry into the sacrum. At insertion of the injection needle, but before injection, the patient stated that he felt pain in his penile shaft in the same two regions as during masturbation, indicating that the injection needle was directly stimulating the nerve roots relevant to his pain. Within 1 to 2 hours of the injection, he reported mild leg weakness. Then, he underwent repeat neuro-genital testing

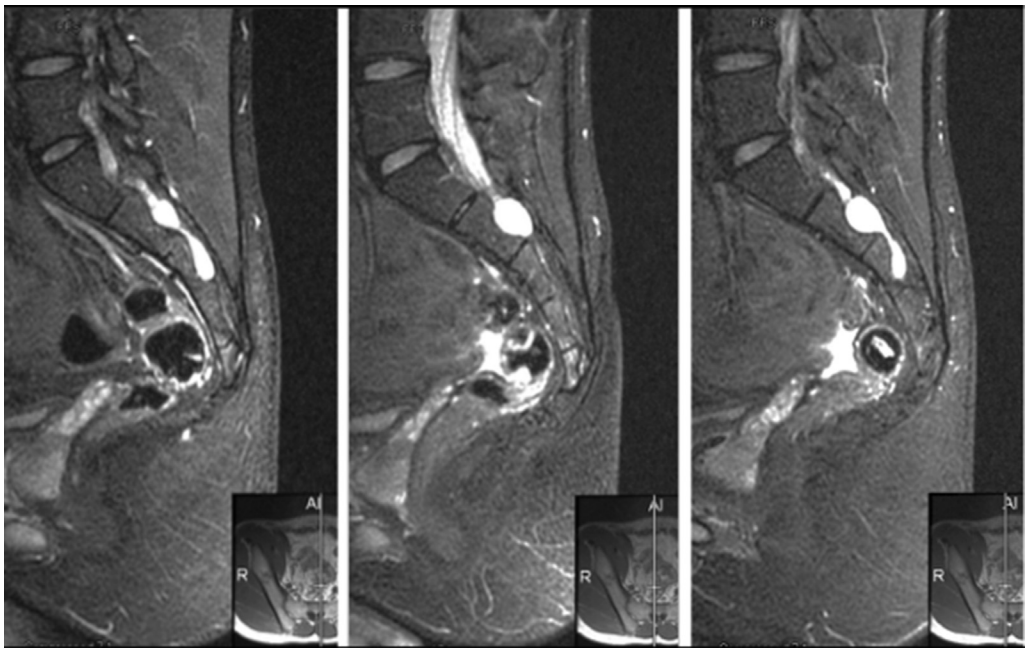

Figure 2. Sagittal views of the patient's multiple Tarlov cysts (white elliptical objects) at sacral levels 2 and 3 in locations relative to the midline that are shown in the corresponding small frontal images at the lower right side of each panel. The cysts appear white in these water-enhanced T2-weighted images because they contain cerebrospinal fluid.

immediately after the S2 and S3 local anesthetic administration. He reported decreased sensation of urine flow through the urethra. Then, he reported that compared with control values, there was decreased perception of pinprick and of hot and cold based on threshold testing at the following dermatomal levels: buttocks near the anus (S1—S4), at the back of the thighs (S1 and S2), and the back of the calves (S1 and S2). Then, he was asked to masturbate in the clinic. His severe penile pain onset with masturbation was delayed. The bupivacaine dose was intentionally low to avoid undue diffusion to the cephalad levels. A higher dose might have had a greater effect on attenuating the sexual pain with arousal.

Based on the evidence developed, by the S2 and S3 local anesthesia procedure, that genital sensory sacral spinal nerve root irritation by the Tarlov cysts was generating the penile pain, he underwent a subsequent injection test of intrathecal morphine injection using 0.2 mg of Duramorph 10 mg/10 mL. The morphine solution is prepared by adding 2 mL of Duramorph 10 mg/10 mL to normal saline 18 mL and injecting 2 mL of the solution directly to the spinal cord in the subarachnoid space at the L1 and L2 vertebral levels. This is the location of the conus medullaris, where the S2 to S4 sensory sacral spinal nerve roots undergo their first synapse in the spinal cord. The rationale for injecting morphine, rather than bupivacaine, was that morphine acts on the spinal cord to induce analgesia but not anesthesia,[20] attenuating pain while still allowing for the perception of nonpainful tactile stimulation.

Within 1.5 hours of the intrathecal morphine injection, the patient returned to the sexual medicine clinic and masturbated again. This time he reported that he had neither penile pain nor penile numbness during the masturbation and could ejaculate within minutes. Normally, the pain would start within seconds of initiating masturbation and the intensity of the pain would compel him to stop stimulation. However, he did feel pain from the pelvic floor muscle contraction at orgasm. He underwent repeat neuro-genital testing after the intrathecal morphine administration.[16,17] He reported decreased sensitivity to pinprick and decreased hot and cold perception in response to heat applied to the buttocks (S1—S4), posterior thighs (S1 and S2), and posterior calves (S1 and S2) compared with the control site and a diminution in the sensation of urine flow. In addition, he noted that placement of a catheter into the urethra to assess its sensitivity induced a sensation of pressure, but not pain.

All these patient observations and neuro-genital testing findings were consistent with a radiculopathy of the genital sensory sacral spinal nerve roots that likely resulted from the Tarlov cysts. The hypothesized diagnosis was Tarlov cyst-mediated neuropathy of the sensory pelvic nerve.

The patient decided to undergo definitive surgical intervention of the Tarlov cysts. The surgical procedure to resolve the Tarlov cysts involved laminectomy, exposure of the Tarlov cysts, sparing of aberrant sensory nerve fibers in the cyst wall, cyst wall resection, and imbrication.[12] Tarlov cysts form at the transitional zone between the dura mater and the more fragile perineurium just distal to the dorsal root ganglia.[21] His Tarlov cysts had already eroded the bone of his sacrum, a typical significant pathology caused by the pressurized cyst filled with cerebrospinal fluid exerting chronic pressure against the bone. At the time of surgery, it was observed that the cause of the sacral spinal nerve root compression and symptoms of sacral radiculopathy were

apparently the large, intrasacral meningeal cysts in the sacrospinal canal, causing compression of the adjacent sacral spinal nerve roots, in particular the S2 and S4 nerve roots.[21]

The patient's postsurgical recovery followed a sequence of gradual but steady improvements. Twelve months after surgery he reported experiencing painless and even pleasurable ejaculatory orgasms during partnered sexual intercourse. From the diary maintained by him and his family (Table 2), within the first week after surgery, the pain he usually experienced with sexual stimulation came on "hard" at 45 seconds vs his preoperative experience of 15 seconds. By 6 weeks, the pain was delayed to 3 minutes, with a more gradual onset and less intensity. At 5 months, he could undergo sexual stimulation for 5 minutes before initiation of pain and could ejaculate for the first time in his life "while not blocked," although the ejaculation was "extremely painful." Over the 6 to 12 months postoperatively, pain at ejaculation became less and less, and there was no penile pain with masturbation. By 1 year postoperatively, he reported "hypersensitivity to the point of difficulty differentiating between pain and hypersensitivity." By 13 months postoperatively, the hypersensitivity had disappeared, and he "started to feel pleasurable sensation," which by 14 months was increasing, although still accompanied by hypersensitivity at the tip of the penis.

At the last review 18 months postoperatively, the patient had been in a 7-month sexual relationship and was able to experience sexual intercourse, including ejaculation, without pain. He completed sexual function questionnaires that were compared to preoperative values (Table 1). Occasionally he noted an "increased sensitivity" at ejaculation, but because he had nothing with which to compare it, this could be within the normal range. He is proceeding with his life and school, typical for his age, with his mental well-being established, thus avoiding a potentially life-threatening depression.

Table 2. Summary of diary excerpts maintained by the patient and his family

Time after surgery	Sexual health effect
1 wk	Pain came on "hard" at 45 s (typically came at 15 s before surgery)
	Pain built more gradually than before surgery
6 wk	Pain on masturbation started at ~3 min
	Pain more gradual, level of pain not as intense
5 mo	Pain started at 5 min but able to work through pain
	Pain considerably less than before surgery—able to "finish" (ejaculate) for first time in life while not blocked
	Ejaculation extremely painful
6–12 mo	No pain during masturbation
	Pain became less and less at ejaculation
1 y	Hypersensitivity to point of difficulty differentiating between pain and hypersensitivity
13 mo	Hypersensitivity during ejaculation gone
	Started to feel pleasurable sensation
14 mo	Pleasurable sensation increasing
	Hypersensitivity at tip of penis
18 mo	Pleasurable sexual activity
	Hypersensitivity gone

DISCUSSION

This case report describes an unusually complex sexual dysfunction that was refractory to management for more than 7 years but was eventually successfully managed by multiple health care providers of various disciplines.

It is evident that a single health care provider cannot be expected to possess all the skills and knowledge needed to have formulated, tested, and proved the eventual diagnosis of Tarlov cyst-mediated radiculopathy of the afferent sensory sacral spinal nerve root causing neuropathy of the pelvic nerve rather than the pudendal nerve.

One of the aims of this case report is to describe the benefits of a novel collaborative association among three specialties: sexual medicine, neurophysiology, and spine surgery. This collaboration allowed us to "bring to the table" multiple critical skills that any single provider would be unable to possess. This collaboration allowed a patient with puzzling, complicated set of sexual medicine complaints to undergo a logical and rational management plan that resulted in suspecting that sacral spinal nerve root compression was the etiology of the neurogenic sexual dysfunction and directing surgical decompression of the suspected irritated radiculitis of the sacral spinal nerve root. In the meantime, we have used our collaborative protocol to successfully restore sexual health in multiple other patients with suspected irritated or compressed sacral spinal nerve root. We will soon report on these cases.

The current protocol for this collaborative association is shown in Figure 3. First, the sexual medicine physician should identify the specific sexual problem through a detailed biopsychosocial sexual history, physical examination, and laboratory tests.

Second, the sexual medicine physician should perform specific neuro-genital diagnostic testing, including such procedures as (i) genital biothesiometry (Figure 4A), (ii) genital temperature perception testing (Figure 4B), (iii) sacral dermatome testing (Figure 4C), and (iv) bulbocavernosus reflex latency testing.[16,17] In this patient's case, only the first three tests were performed, because bulbocavernosus reflex latency testing was not yet available. Instead, urethral distension testing was performed as a neuro-genital test. If these neuro-genital test results are abnormal, they provide evidence to suspect S2 to S4 neurologic pathophysiology.

Third, MRI of the sacral, not pelvic, and lumbar spine should be performed to identify whether there is spinal pathology and, if so, whether it is potentially treatable. These pathologies can include spinal meningeal cysts such as Tarlov cyst(s), disc impingement, annular tears, facet (spinal synovial) cysts, and/or spinal stenosis.[21,22] A sacral MRI specifically visualizes the

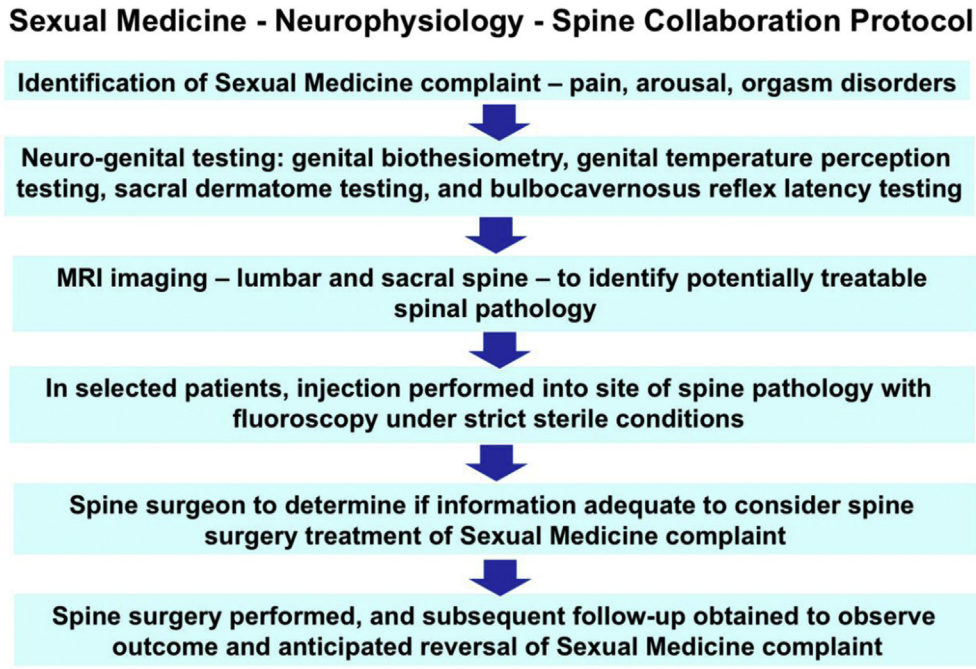

Figure 3. Collaborative protocol used to restore sexual health in patients with sexual dysfunctions from suspected radiculitis from irritated or compressed sacral spinal nerve roots.

anatomic orientation of the nerve roots in relation to the sacrum. The goal is to obtain imaging sequences that identify the anatomic characteristics of the Tarlov cyst. The non-specific pelvic MRI often will have non-orthogonal sequences that make it difficult to characterize the Tarlov cyst.

Fourth, if the sexual medicine disorder is a complaint based on nerve hyper-reactivity (eg, PGAD) or a sexual pain condition worsened during arousal or orgasm (as with this patient), it is advisable to perform an epidural and/or intrathecal injection under fluoroscopy under strict sterile conditions with administration of local anesthetic into the site of the neurologic pathology or of morphine at the conus medullaris, respectively, the latter of which is the site of the first synapse of the pudendal and pelvic nerves. This local spinal anesthesia and/or analgesia resulting in a decrease of the sexual symptoms should serve as evidence that the symptoms are a neuropathy, with a likely relation with the spinal or more peripheral neuropathy. Local anesthesia injections of regions near the peripheral branches (dorsal, perineal, and inferior rectal) of the pudendal nerve or the pudendal nerve itself should be performed preliminarily and fail to eliminate the PGAD or pain complaint, thereby eliminating the possibility that the pathology is in or near the genital tissue per se vs radiculitis in the sacral spinal nerve roots.

Fifth, spine surgery should be considered, assuming the collaborating specialists agree that the pathophysiology is related to a treatable spinal pathology.

Sixth, after surgery is performed, the patient should be followed for at least 1 year to observe whether the suspected neurologic injury is reversed and sexual function is restored. As routinely practiced in sexual medicine, the patient completed a number of validated questionnaires before and after treatment. The findings are presented in Table 1. Using the McGill Pain Questionnaire,[23] the patient rated his pain intensity as 5 of 5 before his surgery and as 0 of 5 one year after surgery. In addition, he was asked to mark his pain along a continuum ranging from "no pain" to "worst pain." Before treatment the patient checked the "worst pain" box and 1 year after treatment he chose "no pain." The International Index of Erectile Function,[24] a tool that uses 15 questions assessing the five domains of erectile and sexual function, showed clinically meaningful improvement before and after treatment. The patient had marked improvement in erectile function, orgasm function, intercourse satisfaction, and overall satisfaction. The only dimension that remained unchanged was the patient's sexual desire, which was not affected by his pain condition. The patient also showed substantial decrease in sexual distress as noted on the scores of the Sexual Distress Scale—Revised[25] and a decrease in the Perceived Stress Scale.[26] Symptoms of depression also were decreased as shown in the Perceived Health Questionnaire—9 score.[27]

The hypothesis is that men and women with suspected neurologic-based sexual health concerns can realize, as did this patient, the benefit of a novel collaboration among colleagues with special interests in neurologic disease and that this partnership enabled restoration of mental and physical sexual health.

It was the opinion of the collaborative group that the four sacral Tarlov cysts (right and left S2 and S3) composed the critical etiologic factor adversely affecting the afferent component of the pelvic nerve underlying the symptoms of severe pain with

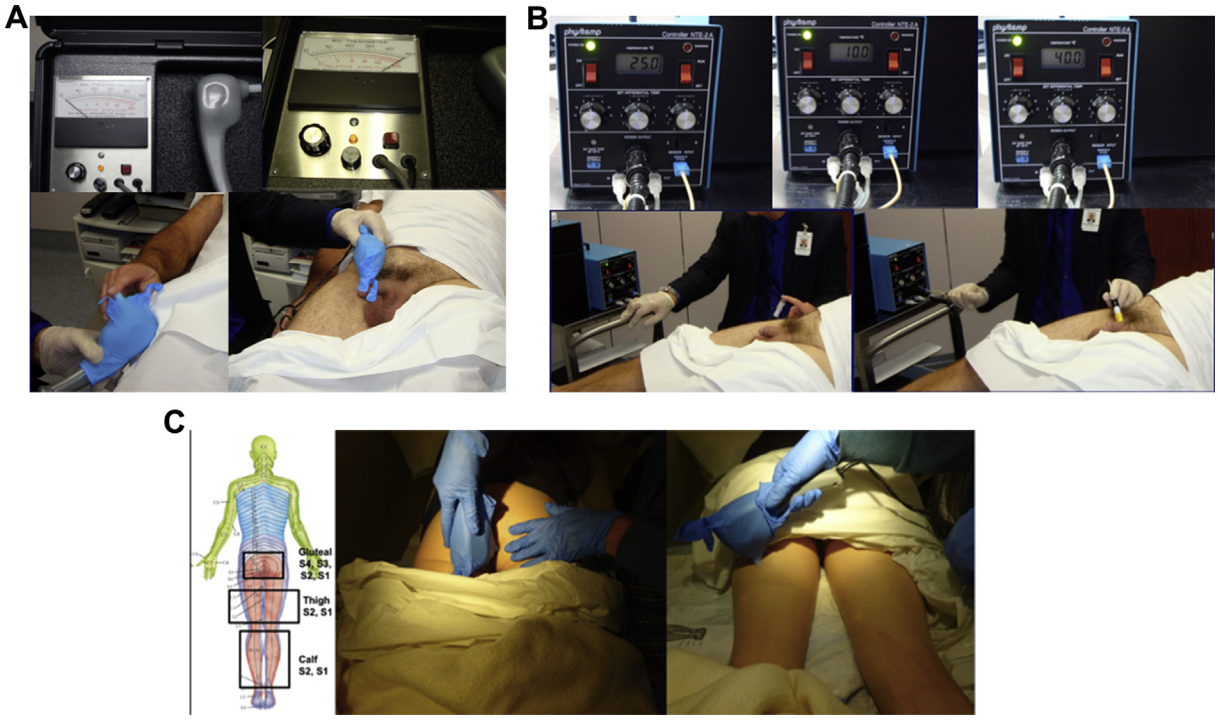

Figure 4. Panel A shows neuro-genital testing using penile biothesiometry. The non-genital site for determination of vibration perception is the pulp of the index finger. Genital sites for determination of vibration perception include the glans penis and right and left sides of the penile shaft. Panel B shows neuro-genital testing using penile temperature perception testing. The non-genital site is the pulp of the index finger. Genital sites include the glans penis and right and left sides of the penile shaft. Panel C shows neuro-genital testing using sacral dermatome testing. The non-genital site for determination of vibration perception is the pulp of the index finger. Genital sites for determination of vibration perception include the right and left gluteal area, right and left side posterior thigh, and right and left posterior calf region.

initial onset of penile erection for sexual activity. This was based on the following facts: (i) the nature of the sensory stimulation that induced his pain was likely related to neuropathy of the afferent component of the pelvic nerve that enters the spine at S2 to S4, (ii) the location of the pain was in the penis (S2—S4), (iii) pudendal nerve blocks failed to affect the severe penile pain during sexual arousal, (iv) there was lack of efficacy of the plethora of prescribed medications, (v) four Tarlov cysts on the genital sensory nerve roots were present, and (vi) there was anecdotal evidence that Tarlov cysts can be sequelae to physical trauma of the pelvic region,[22] which was plausible in his history after a fall onto the "monkey bars" as a young child.

The persistence of the pain despite the local anesthetization of the penile skin, and despite the dorsal nerve and pudendal nerve blocks, provided further evidence that it was the pelvic nerve, that conveyed the pain signal in this case. The dorsal nerve of the penis, a component of the pudendal nerve, conveys sensory activity from the penile skin. The pelvic nerve, whose efferent (parasympathetic) component innervates the corpora cavernosa to produce penile erection, also contains a sensory afferent branch from the deep tissues of the penis,[19] most likely from the corpora cavernosa. This could account for the finding that the local anesthetic, lidocaine, injected into the corpora cavernosa produced general penile numbness, blocking penile sensitivity and pain of masturbation. It is consistent with a report by Komisaruk et al,[28] in a mapping study using functional MRI, that *mild* stimulation of the penile shaft activated a region of the genital sensory cortex (the paracentral lobule) different from that activated by *squeezing* the penile shaft, suggesting innervation by two different penile sensory nerves: pudendal and pelvic, respectively.

Although there is scant literature on the existence and function of penile pelvic nerve afferents compared with the literature on pudendal nerve afferents, the present observations provide evidence that pelvic nerve afferents are at least as important in the normal elicitation of orgasm and ejaculation as pudendal nerve afferents.

Tarlov cysts contain aberrant sensory fibers in the cyst wall and/or within the cyst, and the high-pressure cerebrospinal fluid that fills the cysts impacts them against the surrounding bone of the sacrum, generating mechanical abrasion of the nerve fibers. The best evidence that these cysts can produce peripheral neuropathy is the elimination of this patient's intense pain of orgasm and ejaculation as a result of the surgery relieving the physical abrasion produced by the Tarlov cysts. However, there is a prevalent belief among health care providers that Tarlov cysts are asymptomatic.[29,30] In radiology reports, the possible significance of Tarlov cysts is often dismissed or their existence is not even reported. Two recent reports have provided evidence that they can play a significant role in generating genital neuropathology,

discomfort, and pain. Twelve of 18 women who complained of PGAD, and who submitted their MRI records for evaluation, were reported to have at least one Tarlov cyst at S2 to S4,[8] and definitive surgical management of Tarlov cysts at the sacral level decreased or eliminated PGAD symptoms in 10 of 11 women.[12]

The present case study provides evidence that Tarlov cysts can generate significant neuropathology in man. The widespread but mistaken attitude that Tarlov cysts are asymptomatic has likely discouraged research on their effects. Acute or chronic genital sensory nerve root irritation produced by Tarlov cysts could produce excessive stimulation, resulting in hypersensitivity and/or pain (hyperfunction), whereas intense and/or chronic pressure on the roots could eventually compromise nerve function, leading to sensory deficits (hypofunction). Thus, it would seem advisable to assess, or at least to rule out, the possible role of Tarlov cysts as a possible etiology in various sexual dysfunctions in men, including (i) painful erection and/or painful ejaculation, (ii) arousal disorders such as recurrent priapism or PGAD, and (iii) orgasm disorders such as anorgasmia, muted lowered intensity of orgasm, delayed orgasm, or pleasure dissociative orgasmic dysfunction.

In summary, this case report stresses that in the management of neurogenic sexual health concerns, Tarlov cysts can be a critical etiologic factor adversely affecting the sensory component of the pelvic nerves, resulting in symptoms of severe pain with initial onset of penile erection for sexual activity, and can produce extremely painful penile neuropathy. This young man realized postoperative restorations of his sexual life and his mental well-being. Furthermore, this case report underscores the benefits of a novel collaborative protocol among sexual medicine, neurophysiology, and spinal surgery specialties, which can provide a rational-based process of care for patients of sexual medicine. The recent experience of this collaborative group, exemplified by the present case study, emphasizes the potential benefit of considering a diagnosis of radiculopathy of the sensory sacral spinal root resulting in a wide array of disorders of the sexual system in men and women.

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