Persistent genital arousal disorder: a special sense neuropathy

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Abstract

Introduction: Persistent genital arousal (PGAD) is a syndrome of unprovoked sexual arousal/ orgasm of uncertain cause primarily reported in female patients. Most patients are referred for mental-health treatment, but as research suggests associations with neurological symptoms and conditions, there is need to analyze cases comprehensively evaluated by neurologists.

Methods: The IRB waived consent requirements for this retrospective university-hospital study. We extracted and analyzed neurological symptoms, test, and treatment results from all qualifying participants’ records and recontacted some for details.

Results: All 10 participants were female; their PGAD symptoms began between ages 11 to 70 years. Two patterns emerged: 80% reported daily out-of-context sexual arousal episodes (=30/day) that usually included orgasm and 40% reported lesser, often longer-lasting, nonorgasmic arousals. Most also had symptoms consistent with sacral neuropathy—70% had urologic complaints and 60% had neuropathic perineal or buttock pain. In 90% of patients, diagnostic testing identified anatomically appropriate and plausibly causal neuronal lesions. Sacral dorsal-root Tarlov cysts were most common (in 4), then sensory polyneuropathy (2). One had spina bifida occulta and another drug-withdrawal effect as apparently causal; lumbar disc herniation was suspected in another. Neurological treatments cured or significantly improved PGAD symptoms in 4/5 patients, including 2 cures.

Conclusions: Although limited by small size and referral bias to neurologists, this series strengthens associations with Tarlov cysts and sensory polyneuropathy and suggests new ones. We hypothesize that many cases of PGAD are caused by unprovoked firing of C-fibers in the regional special sensory neurons that subserve sexual arousal. Some PGAD symptoms may share pathophysiological mechanisms with neuropathic pain and itch.

Keywords: Neuropathic pain, Pelvic pain, Tarlov cysts, Peripheral neuropathy, Spinal cord, C-fibers

1. Introduction

The anatomy and physiology—and thus the innervation—of sexual arousal are dimorphic, but it has been studied almost exclusively in male patients, and the peripheral and spinal pathways and neurotransmitters mapped primarily in rodents.15,16,27 Studies mapping human arousal are rare and mostly conducted in spinal cord–injured or multiple sclerosis patients.1,20 Veterans Administration and other investigators have studied effects of myelopathies, radiculopathies, neuropathies, and various medications on male arousal,20 but research in female patients is nearly nonexistent. Women’s complaints of inappropriate arousal are typically attributed (by predominantly male evaluators) to psychopathology or misinterpreted as beneficial.4

Here, we begin neurological investigation of persistent genital arousal disorder (PGAD), a largely female-reported syndrome of out-of-context sexual arousal and/or orgasm. PGAD has been mostly investigated by psychologists. With physicians and neuroscientists largely unaware of it, medical causality has not been systematically investigated.14 Feigenbaum and Komisaruk established the firmest association to date, with sacral Tarlov cysts. These form exclusively on and can damage sensory ganglia and roots.2,11 Some cases are attributed to brain effects of serotonergic and dopaminergic drugs,5,22 and sexologists have hypothesized that other neurological problems may be associated, mentioning restless leg syndrome, fibromyalgia, genital sensory hyperesthesia, neuropathic pain, and sensory neuropathy, but we are unaware of previous neurologically focused investigations.7,8,17,19,22–24,26
2. Methods

A lack of standardized nomenclature (synonyms include persistent sexual arousal syndrome and restless genital syndrome) and billing codes precluded systematic case ascertainment, so we reviewed records from our university–hospital neurology practices for PGAD mentions and solicited additional referrals regardless of whether neurological symptoms were present. The review board waived consent, although we obtained verbal consent to anonymous publication. All genders and ages were eligible; inclusion required neurological evaluation of diagnosed or suspected PGAD, and some patients were reinterviewed. We analyzed demographics, medical histories and examinations, results about localization, etiology, and treatment.

3. Results

All participants were female, and on average 53.4 years old on December 31, 2018 (Table 1). Ages at PGAD onset ranged from puberty to postmenopausal. We identified 2 patterns of arousal—episodic and sustained. Eighty percent of patients reported daily transient sexual arousals (minutes/few hours) with 40% reporting longer, lesser near-continuous arousals for days-years (2 had both). All PGAD illnesses began as anorgasmic but almost always progressed to include spontaneous orgasms. Patient 4, with ≤30 arousals daily, had 2 unprovoked orgasms in front of a hospital teaching-conference audience. Almost all patients tried masturbation to terminate arousals, and this helped 20%. Patient 10 masturbated 4 to 5 times daily despite the lack of pleasure, to obtain a few hours relief. Patient 3 induced several orgasms each afternoon to quell symptoms until the next morning. Five reported no postorgasm refractory relief, and patient 6 avoided all vulvar contact because of allodynia.

Chronic PGAD always terminated sexual relations. All 6 partnered patients initially sought sex during their arousals, but all of their partners came to perceive their approaches as too frequent and/or “mechanical,” and terminated sexual relations, although all marriages continued. Among the 3 patients who were virgins at PGAD onset, 2 remained abstinent and 1 tried intercourse only once, an encounter abrogated by vulvodynia. Every patient reported that PGAD caused new or worse depression and anxiety. Onset in childhood was bewildering, causing confusion, shame, and fear. All patients considered themselves disabled from PGAD and associated symptoms, and most had curtailed daily activities. At presentation, only 20% of patients’ physicians recognized their symptoms as PGAD, so most self-diagnosed online. Before onset, all were functioning well and none had major psychiatric diagnoses, yet several reported psychiatric attribution and treatment—for example, with sex therapy and electroconvulsive treatments.

Eighty percent of patients first sought care for their other pelvic symptoms and only mentioned PGAD after establishing trust. Among medical consultations, neurological evaluations were the most productive. They documented colocalizing somatosensory symptoms in 90%, including perineal, buttock, or leg pain and/or sensory loss. Neurological testing was also productive, with 78% (6/8) of sacral magnetic resonance imaging studies revealing radiculopathy, 2/2 nerve-conduction studies diagnostic for sensory polyneuropathy along with 2/5 lower-leg, PGP9.5-immunolabeled skin biopsies. Among 2 composite autonomic function tests, 1 was abnormal, the other borderline. Abnormal urodynamic and anorectal manometry testing confirmed myelopathy in the spina bifida patient. Four electroencephalograms in 2 patients were unremarkable, including one capturing 4 spontaneous orgasms.9

Psychiatric treatment was universally ineffective, including 7 psychiatric hospitalizations and 17 electroconvulsive therapy sessions for patient 10. Gynecological and urological treatments, including medications, injections, and electrotherapies, were also ineffective. Local anesthetics and/or corticosteroid injections never had lasting benefit, but a few gave temporary relief, suggesting the potential for diagnostic localization of hyperexcitable sensory nerves as with neuropathic pain conditions. Genitofemoral nerve blocks gave transient relief to 2/3, but pudendal nerve blocks were ineffective or worsened symptoms in 5/5. All epidural corticosteroid injections worsened symptoms. One intravaginal botulinum toxin administration was ineffective.

By contrast, neurological treatment was effective in 80% of patients. Gradual duloxetine taper (patient 5) and Tarlov-cyst resection (patient 2; Fig. 1) were curative. Immunoglobulins (2 grams/kg/4 weeks) improved patient 8’s PGAD and motor symptoms dramatically. Another Tarlov-cyst patient found intrathecal pressure reduction helpful, whereas surgical resection was ineffective for another.

4. Discussion

This report associates PGAD with disorders and lesions of the lower spinal cord, roots, and nerves that control sexual arousal and orgasm. Genital sensory innervation is mostly through the dorsal nerve of the clitoris/penis, a branch of the pudendal nerve that enters the cord through S4 dorsal roots to excite T12-S1 dorsal-horn interneurons (laminae VI and X27)1,16 and send axons up the dorsal columns and gracile fasciculus to affect the brain widely.3,12 In female rats, electrophysiological recordings link pelvic contractions to rhythmic pudendal nerve firing, with L4 spinal cord injury increasing this firing.1 Autonomic mapping in mice identifies hypogastric sympathetic afferents entering at L2,16 with efferents exiting the T12-L2 ventral roots and white rami to synapse paravertebally then send postganglionic fibers through gray rami to the hypogastric nerve. Parasympathetic efferents arise from S2-5, exit as splanchnic nerves through the inferior hypogastric plexus to synapse in ganglia in pelvic organ walls, and increase pelvic blood flow and other arousal responses.

Given our patients’ lesion localizations, etiologies, and colocalizing neurological signs and symptoms, we propose that at least some PGAD cases arise from lesions affecting the sacral sensory networks that transmit sexual arousal—that it is a disorder of special sensation akin to neuropathic pain and itch. To reflect this, we propose congruent Greek-derived neurophysiologic nomenclature. For sexual arousal after nonsexual stimulation, we suggest “allodygeria” (allo/διγερo) for “other” plus diegeria/διγερησις (sexual arousal), analogous to “allodynia” for pain and “alloknesis” for itch. For spontaneous sexual arousal or orgasm without physical or mental stimulation, we propose “allodiogeneis” from afformato/αλλομετα (unprovoked) and diegerias/διγερησις (sexual arousal). A genital-pelvic nomenclature has been developed,19 but adding conventional neurological nomenclature could improve general medical awareness, care, and research.

Our findings will have clinical implications if confirmed because most PGAD patients now linger medically undiagnosed and untreated. Patient-initiated internet sites document thousands of (usually female) questioners. Therefore, this small series, although among the largest of examined patients, cannot fully represent PGAD nor provide accurate prevalences, causes, or treatment outcomes because of referral bias. However, it offers an interim clinical framework. It independently identifies sacral dorsal root Tarlov cysts as causa2,11 and strengthens associations with
Initially once every 3 spontaneous orgasms

40% 1–2 hours of near-continuous strong arousal per day, 60% 1–2/day arousal without orgasm, severe episodes of mechanical continuous mild or strong arousal, 1 spontaneous orgasm/mo while sleeping

Table 1

| Patient characteristics. | Patient 1 | Patient 2 | Patient 3 | Patient 4 | Patient 5 | Patient 6 | Patient 7 | Patient 8 | Patient 9 | Patient 10 |
|--------------------------|-----------|-----------|-----------|-----------|-----------|-----------|-----------|-----------|-----------|-----------|
| Demographics             | Caucasian female age 29.3 years | Arabic female age 35.9 years | Caucasian female age 36.8 years | Caucasian female age 42.1 years | Caucasian female age 50.0 years | Caucasian female age 58.4 years | Caucasian female age 59.0 years | Caucasian female age 60.9 years | Caucasian female age 71.8 years | Caucasian female age 79.8 years |
| Onset                    | At menarche (age 12), developed unprovoked arousals without orgasm | At 70, developed multiple (6) daily episodes often including orgasm, worse in afternoon. Gradual lessening after adolescence | At 32, developed unprovoked arousals without orgasm | At 30, near menarche, developed unprovoked arousals without orgasm | At 46, developed unprovoked arousals without orgasm for 2 days, then with orgasm. | At 50, developed unprovoked arousals without orgasm | At 58, developed continuous strong orgasms for 2–3 months, then with orgasm | At 53, developed continuous low-level arousal without orgasm | At 69, developed unprovoked clitoral tingling and arousal, then orgasms early on | At 70, developed unprovoked arousals without orgasm |
| Temporal characteristics of PGAD episodes | Multiple (≤10) daily episodes of arousal for 5–40 minutes, 20% include orgasms that gave a few hours relief | Multiple (≤5) daily episodes that never caused orgasm | Multiple (≤5) daily episodes often including orgasm, worse in afternoon. Near-continuous arousal without orgasm, severe mechanical vulvodynia | Multiple (≤4) daily brief episodes of almost immediate orgasm even during sleep | Multiple 2 hours continuous strong arousal during sleep, severe mechanical vulvodynia | Near-continuous arousal without orgasm, severe mechanical vulvodynia | Near-continuous arousal without orgasm, severe mechanical vulvodynia | Near-continuous arousal without orgasm, severe mechanical vulvodynia | Near-continuous arousal without orgasm, severe mechanical vulvodynia | Near-continuous arousal without orgasm, severe mechanical vulvodynia |
| Precipitants             | Sitting, vibration travel (sitting + vibration) | Standing, lying flat, menses | Premenses and menses | Sitting, squeezing legs together | Sitting, vulvar contact including clothing gives sensation of vaginal distention | Sitting, vibration (sitting + sitting) | Sitting, travel (vibration + sitting) | Sitting, vulvar contact including clothing gives sensation of vaginal distention | Sitting, travel (vibration + sitting) | Sitting, travel (vibration + sitting) |
| Ameliorants              | Avoiding precipitants | Normal aging | None | Restarting duloxetine, cooling vulva, drinking wine | Wearing panty liners to prevent brushing against vulva, avoiding pants with seams | Avoiding some positions | None | Sitting, lying down | None | None |
| Effects of masturbation  | Tried, not helpful | Tried, not helpful | Tried, used daily afternoon masturbation with several orgasms to dampen baseline arousal until next day | Tried, not helpful | Tried, only brief refractory period, so not helpful | Tried, only brief refractory period, so not helpful | Tried, only brief refractory period, so not helpful | Tried, only brief refractory period, so not helpful | Tried, only brief refractory period, so not helpful | Tried, anhedonic masturbation 4–5× daily gave 1 hour of relief |
| Urologic symptoms        | Neurogenic bladder since infancy, self-catheterizes | Frequency, urgency, mild hesitancy, sense of incomplete voiding | Hesitancy, some incontinence | Strong urgency during and between episodes | Urgency, frequency, urinary incontinence | None | Frequency (15× daily), occasional bladder pain labeled interstitial cystitis | None | None | None |
| Other neurologic signs and symptoms | Sacral dimple at birth, neurogenic bladder, bowel since birth, use of cecostomy tube to void bowels, perineal sensory loss, pain radiating to upper inner thighs, rarely to toes | Age 23 neuropathic pain in left vulva, rectum, upper thigh, bilateral low back and leg pain, radicular sensory loss left vulva, buttock, thigh, chronic occipital headaches | Tachycardia, gastrointestinal dysmotility with nausea, vomiting, 50 lb weight loss, neurogenic constipation since childhood | Restless leg movements at night and white sleeping | Perineal and right buttock burning, itching, pain, severe pain with vaginal insertion | Low back, right leg pain and numbness, occasional clitoral pain, alldynia | Neuropathic pain, numbness, itch in hands and feet, postorgasm headache attributed to CSF leak from cervical TC, chronic fatigue, exertional intolerance, syncope, abnormal GI motility | Pelvic pain soon after onset at right T11–T12, mons-buzzing, poking feelings. Pin examination of right labium majus felt as not sharp, tingling, “sexy” | Bilateral paresthesias in thighs and legs, right leg cramp |

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|-----------|-----------|-----------|-----------|-----------|-----------|-----------|-----------|-----------|-----------|
| Obstetric-gynecology history | Nulliparous, severe pelvic pain worse on right, severe vaginal insinational pain, polycystic ovarian syndrome | Nulliparous, bilateral ovarian cysts | Nulliparous, extraterine endometriomas and adhesions, managed hormonally, surgically | Nulliparous, menorrhagia-treated hormonally and surgically; previously anorgasmic despite attempts with stimulator | Three uncomplicated vaginal childbirths; premenopausal menorrhagia | One pregnancy terminated, premenopausal menorrhagia | Two pregnancies, one preterm, one twin | One pregnancy; Cesarian delivery; endometriosis treated with lysis of adhesions; uterine fibroid; genital herpes since youth | Tubal ligation, breast biopsy, abdominal surgery for ovarian cyst and appendectomy | Two uncomplicated vaginal childbirths |
| Sexual sequelae | No sexual experience at PGAD onset, one failed intercourse attempt, then abstinent | No sexual experience at PGAD onset, remained abstinent | Sexual relations with fiancé ended within a year, then abstinent | Remained abstinent until PGAD resolution | Sexual relations with husband ended; then abstinent | Sexual relations with husband ended; then abstinent | New near-immediate orgasm during sex, sexual relations with husband ended; then abstinent | Clitoral stimulation painful, sexual relations with husband ended; then abstinent | Sexual relations with husband ended; then abstinent |
| Psychiatric symptoms | Depression, anxiety | Depression, anxiety | Depression | Depression, anxiety | Depression, anxiety | Depression, anxiety | Pre-existing depression | Depression | Depression |
| Initial PGAD attribution | Attributed to STD despite no previous partners | Depression, stress, myofascial leg pain plus occipital neuralgia | Uncertain | Hypersexuality, seizures, endocrine dysfunction | Sudden duloxetine discontinuation | Sexologist-diagnosed PGAD, uncertain of cause | Prolonged sitting position during medical procedure | No explanation offered by gynecologist | Pelvic neuropathy |
| Lumbosacral MRI (Fig. 1) | Malfusion of S1 arch, L4-S1 facet arthropathy, L5-S1 HNP with mild left root compression | Sacral TC; 6.3 cm at S1, into pelvis compressing illopoas, bilateral S2 cysts with foraminal erosion; mild lumbar DJD, left S2 | L5-S1 HNP with moderate foraminal stenosis, right L5 root contact | Noncontributory, sacral MRI with only mild sacroiliac DJD, lumbar mild DJD, L45, L5-S1 facet arthropathy, bilateral mild–moderate foraminal stenosis | Sacral TC; small right S1, large bilateral S2, S3 small S4 | Noncontributory, sacral MRI with only mild sacroiliac DJD, lumbar mild DJD, L45, L5-S1 facet arthropathy, bilateral mild–moderate foraminal stenosis | Lumbal TC, bilateral L5 1 cm on right, two 0.4-cm cysts on left | Sacral TC; 5-mm right S3, L4-L5, L5-S1 DJD, disc bulges, bilateral mild–moderate foraminal stenosis | Sacral TC; large bilateral S3, S4 |
| Peripheral nerve testing | Urodynamics with outlet obstruction, postvoid residual, anorectal manometry with high resting anal pressure with outlet obstruction, low squeeze pressure, poor rectal sensation | Abnormal lower leg skin biopsy (103 ENF/mm² skin surface area, at ≤1st centile of predicted), borderline composite autonomic function testing | Normal lower leg skin biopsy (398 ENF/mm² of skin surface area, at 84th centile of predicted) | Not performed | NCS with right–only slow pudendal nerve motor latency, EMG normal | Normal lower leg skin biopsy (278 ENF/mm² skin surface area, at 86th centile of predicted), EEG noncontributory | In 2015 had 28 ENF/mm² skin surface area; <1st centile of predicted, EMG/NCS with demyelinating + axonal motor sensory changes, normal AFT. 2018 skin biopsies: diagnostic for SFN at distal leg and thigh, reduced sweat-gland innervation at distal leg, AFT diagnostic for SFN | Normal distal leg skin biopsy (255 ENF/mm² skin surface area, at 88th centile of predicted) | Not performed |

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| Patient characteristics | Patient 1 | Patient 2 | Patient 3 | Patient 4 | Patient 5 | Patient 6 | Patient 7 | Patient 8 | Patient 9 | Patient 10 |
|-------------------------|----------|----------|----------|----------|----------|----------|----------|----------|----------|-----------|
| **Other neurologic testing** | Thoracic MRI with T11-12 HNP and mild cord signal changes | None performed | Cervical MRI with mild DJD, 56-panel whole exome sequencing with no pathogenic variants | 3 normal EEGs including 1 during 4 spontaneous orgasms, brain MRI noncontributory | Brain MRI noncontributory | None performed | Cervical MRI with C6, C8 Tarlov cysts, multilevel DJD Brain MRI, PET noncontributory | None performed | Brain MRI, CTA normal; thoracic and lumbar MRI with mild DJD |
| **Current PGAD attribution** | Symptomatic sacral spina bifida occulta | Sacral radiculopathy from multiple Tarlov cysts | Small-fiber neuropathy, possible plexus irritation by endometriomas | LSS1 HNP with L5 radiculopathy | Sudden duloxetine discontinuation | Sacral radiculopathy from multiple Tarlov cysts | Unknown, sacral MRI recommended | Atypical CIDP with small-fiber involvement plus lumbar Tarlov cysts | Sacral radiculopathy from right S3 Tarlov cyst | Sacral radiculopathy from multiple Tarlov cysts |
| **Ineffective symptom treatments** | Pudendal nerve block, empiric anti-infectives for STDs, vaginal Botox injections to bladder neck, clitoris, pelvic floor, topical lidocaine | None offered | Norbttropline offered but declined | Sex therapy, levetiracetam | Unknown | Pudendal nerve blocks, caudal epidural steroids, testosterone, pelvic PT, TENS, tibial nerve stimulator | Pudendal nerve blocks, epidural steroids, trigger point injections, gabapentin, topical amitriptyline, baclofen, local anesthetics | Unknown | Bilateral pudendal, genitofemoral, ilioinguinal, nerve blocks, gabapentin, pregabalin, sertraline, acupuncture, topical lidocaine, amitriptyline/baclofen, ovarian vein embolization |
| **Effective symptom treatments** | Bilateral genitofemoral nerve block gave 80% relief for 3 days only | Mild improvement with duloxetine, acetazolamide | Gradual improvement with aging | Mild improvement with risperidone | None | Mild help from gabapentin cream, genitofemoral nerve blocks gave few weeks of 85% relief | None | None | None | None |
| **Definitive neurologic treatments** | None tried | Complete remission after surgical Tarlov cyst resection 1.5 years ago | None tried; trial of IVig recommended but declined | None tried; surgical consultation for HNP decompression recommended | 5 year total remission after duloxetine 60 mg resumed on day 3; tapered over 3 weeks | Resection of left and right S2 and right S3 Tarlov cysts was ineffective | None recommended or tried | Mg reduced PGAD days from maximum of 30 days/month to 4 days/month and greatly improved other neuropathy symptoms eg, weakness | None tried; acetazolamide trial recommended | None tried; Tarlov-cyst resection recommended but declined |

**AFT**, autonomic function testing; **CIDP**, chronic inflammatory demyelinating polyneuropathy; **CSF**, cerebrospinal fluid; **DJD**, degenerative disk disease; **ECT**, electroconvulsive therapy; **EEG**, electroencephalogram; **EMG**, electromyography; **ENF**, epidermal nerve fibers; **GI**, gastrointestinal; **HNP**, herniated nucleus pulposus; **LS**, lumbosacral; **MRI**, magnetic resonance imaging; **NCS**, nerve conduction study; **PGAD**, persistent genital arousal disorder; **PT**, physical therapy; **TC**, tarlov cyst; **TENS**, transcutaneous electrical nerve stimulation.
sensory polyneuropathy. It adds another case associated with lumbosacral disc herniation and proposes cauda equina malformation and sensory CIDP as potential new causes. It associates PGAD with abrupt duloxetine withdrawal, given that duloxetine resumption was curative, extending reported associations beyond initiation of libido-promoting drugs (eg, dopaminergics) and abrupt discontinuation of libido-inhibitors (eg, serotonergic antidepressants).\textsuperscript{5,6,8,22} Conceivably, male patients merely seek treatment less often, but we propose 3 biological contributors. A total of 90% to 95% of symptomatic Tarlov-cyst patients are women, because of their thinner meninges and tilted pelvis containing more-vertical nerve roots more exposed to CSF pressure waves. In addition, female patients represent 3/4 of many small-fiber neuropathy cohorts,\textsuperscript{13} and 2/3 of US antidepressant users.\textsuperscript{18} Female patients thus have a higher risk of associated neurologic conditions.

For lesion localization, 3-mm-cut sacral MRI and tests for neuropathy were highly useful. Pudendal nerve conduction should be measured more often, particularly with glove electrodes now standard. Brain MRI and EEG were futile.\textsuperscript{23} Regarding treatment, skilled neurosurgeons report good outcomes for Tarlov-cyst resection.\textsuperscript{2} Medical management of PGAD symptom should include gradual tapering of causal medications, and perhaps considering libido-dampening drugs. For nerve and nerve-root lesions, tricyclics, ion-channel blockers, and anti-epileptics—effective for neuropathic pain and itch—deserve consideration. Neurological evaluation and treatment should precede psychotherapy, electroconvulsive therapy, or clitoridectomy.\textsuperscript{25}

Figure 1. Magnetic resonance imaging identifying sacral Tarlov cysts (patient 1). (A) Sagittal view. Green arrows depict trilobed, multiseptate T2 hyperintense, T1 hypointense nonenhancing Tarlov perineurial cysts in the left L5-S1, left S1-S2, and left S2-S3 neural foramina (right S1-S2 cysts were also present and seen on other images). (B) Coronal view. Green arrows depict multiseptated T2 hyperintense, T1 hypointense perineurial cysts in the same patient. The largest cyst, at left L5-S1, tracks along the course of the L5 nerve root and measures 6 cm in greatest dimension.

Disclosures
The authors have no conflict of interest to declare.

Supported in part by NIH R01NS093653 and K24NS059892 (to A.L.O.) and Sydney R. Baer Jr. Foundation (to B.H.P.).

Presented in abstract form to the 2018 meeting of the International Association for the Study of Pain.

Acknowledgements
The authors appreciate the referral of two patients by neuro-urologist Dr. Elise De and the assistance of Dr. Marinos Dalakas with Greek terminology.

Article history:
Received 7 June 2019
Received in revised form 13 September 2019
Accepted 6 November 2019
Available online 6 January 2020

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