

Prospective Study



Sacral Nerve Stimulation in Patients With Refractory Pudendal Neuralgia

Kai-kai Guo, MS¹, Long Wang, MD¹, Fang Liu, MS², Jie-jie Niu, MS², Chao Wang, MS², Shao-hua You, MD¹, Ze-guo Feng, MD¹, and Gui-jun Lu, MS²

From: ¹Department of Pain Medicine, The First Medical Center of PLA General Hospital, Beijing, China; ²Department of Pain Medicine, Beijing Tsinghua Changgung Hospital, School of Clinical Medicine, Tsinghua University, Beijing, China.

Address Correspondence: Gui-jun Lu, MS
Department of Pain Medicine
Beijing Tsinghua Changgung Hospital School of Clinical Medicine, Tsinghua University
168 Litang Road,
Changping District
Beijing, China
E-mail: luguijun301@163.com

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Background: Pudendal neuralgia (PN) is one of the most common forms of genital pain. Only 42.2% of PN patients respond to the first-line treatment. Novel neuromodulation techniques in the treatment of refractory PN patients are urgently required.

Objectives: The aim of this study was to evaluate the treatment effects and adverse events of sacral nerve stimulation (SNS) for patients with refractory PN.

Study Design: A prospective nonrandomized study.

Setting: This prospective analysis included 33 patients who received the phase II surgical implantation.

Methods: A total of 55 eligible PN patients were recruited for SNS treatment after informed consent, and 33 of 55 patients with a minimum 50% improvement were candidates for surgical implantation. Visual Analog Scale (VAS) scores, Self-rating Anxiety and Depression Scale, Quality of life score (SF-36), and sleep monitoring indicators before and after surgery were used to assess the effects of SNS on patients with refractory PN.

Results: Thirty-three patients were included in the final analysis, involving 24 women and 9 men with a mean age of 49.5 years (26-70 years). There was a favorable decrease in pain severity (VAS scores) from 7.1 ± 1.1 at baseline to 6.1 ± 1.0 on postoperative day 1, and 2.8 ± 0.7 at 1 week, 1.7 ± 0.5 at 1 month, 1.1 ± 0.7 at 6 months, and 1.0 ± 0.6 at 12 months after surgery, respectively ($P < 0.05$). The mean score of each section of SF-36 after SNS was significantly higher than that at baseline ($P < 0.05$). Total sleep time and sleep time in each period were significantly prolonged after SNS implantation compared with that before surgery (6 months vs Pre, total: 5.32 ± 1.49 hours vs 3.66 ± 1.19 hours, deep: 2.52 ± 0.63 hours vs 1.36 ± 0.43 hours, light: 1.78 ± 0.42 hours vs 0.99 ± 0.30 hours, rapid eye movement: 1.41 ± 0.29 hours vs 0.89 ± 0.27 hours, $P < 0.05$). No serious device complications were reported during the follow-up period.

Limitations: Large-scale randomized clinical trials are warranted to evaluate the risk factors for prediction of refractory PN.

Conclusions: These data imply that SNS can have beneficial effects on patients with refractory PN.

Key words: Sacral nerve stimulation, refractory pudendal neuralgia, before-after study in the same patient

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Pudendal neuralgia (PN) is a neuropathy of the pudendal nerve that causes pain (1). PN has an estimated incidence of 1/100,000, and is often

misdiagnosed by many pain physicians. Spinosa et al (2) have reported that through literature review, the incidence of PN in the general population is only

1%, and women are more susceptible than men. Orphanet is a European knowledgebase that provides information on orphan drugs and rare diseases, in which PN is proposed to affect 4% of patients receiving pain counseling, and 3 of 7 men are affected. Most pain physicians believe that the actual prevalence of PN may be much higher than that described in the existing literature.

Health managements of PN mainly include behavioral modifications, pelvic floor physiotherapy, analgesics, pudendal nerve block, injections of botulinum toxin A (in case of muscle spasms), surgical nerve decompression, radiofrequency, and spinal cord stimulation of the conus medullaris (3,4). Medications are considered as a conservative therapy, although some PN patients may not benefit from it and a permanent therapy is required. Pereira et al (5) have reported that 42.2% of PN patients respond to the first-line treatment, but in the nonresponders receiving the first-line treatment, the second-line pharmacological treatment is only effective in 8/26 patients. Transsacral block at S2-S4 results in a pudendal nerve block, which can significantly relieve PN symptoms (6). Surgical decompression is recommended in PN patients who have refractory responses or are unresponsive to pain management, like medications of neuropathic pain, infiltrations, rehabilitation, transcutaneous electrical nerve stimulation, and psychobehavioral approaches, which can effectively release the pudendal nerve from entrapment zones, including the space between the sacrospinatus and sacrotuberous ligaments, obturator internus fascia, and infrapiriformis canal (7). However, about 30% of PN patients rarely benefit from a decompression procedure, with a less than 30% gain on the Visual Analog Scale (VAS) scores at 1 year postoperatively (4). Therefore, novel neuromodulation techniques, based on neuropathic characteristics of the pain in the treatment of refractory PN patients, are urgently required.

Sacral nerve stimulation (SNS) is a minimally invasive mean. The procedure is divided into 2 parts (8). Each patient undergoes a screening test during which a quadripolar self-blocking tined lead was inserted in the S3 foramen through puncture under local anesthesia, and a temporary external pulse generator was used for in vitro testing stimulation for 2-4 weeks (phase I). If the patient demonstrates significant improvement in targeted symptoms, the implantation of a permanent internal pulse generator (IPG) is then performed (phase II). Previous studies (9) have indicated that SNS alleviates refractory voiding dysfunction. SNS has become one of

the most accepted methods of stimulation treatment in the field of functional urology. However, there is little research on the application of SNS in the treatment of PN at present. Siegel et al (10) have demonstrated that SNS effectively reduces the severity and frequency of chronic intractable pelvic pain. Moreover, the efficacy of neuromodulation in the treatment of PN remains controversial owing to the small sample sizes in single-center studies, and requires more clinical evidences. Our study aims to thoroughly assess the efficacy of SNS in relieving pelvic pain in PN patients.

METHODS

Patient Recruitment

This was a prospective nonrandomized study with the approval of the institutional review board for assessing the safety and efficacy of SNS in relieving chronic intractable pelvic and/or urogenital pain. A total of 55 eligible PN patients were recruited for SNS treatment after informed consent, and 33/55 patients with a minimum 50% improvement were candidates for surgical implantation. Therefore, 33 patients were included in the final analysis, involving 24 women and 9 men with a mean age of 49.5 years (26-70 years) (Table 1).

Inclusive and exclusive criteria: PN was diagnosed according to the Nantes criteria (11); chronic neuropathic pain lasting for at least 6 months according to criteria of the Neuropathic Pain Diagnostic Questionnaire; refractory pain after conventional pain management and pudendal nerve decompression surgery performed according to Robert et al's (7) technique; and the minimum VAS scores of 5/10. In addition, patients with severe anxiety and depression were excluded, and patients with infection or malignant tumors in the pelvis and sacrococcyx were contraindications for SNS treatment. Patients unresponsive to the test stimulation were tested again or withdrawn, and those with a less than 50% improvement were also not included in the final analysis.

Baseline Assessment

All included patients underwent the same layer display of the lumbosacral nerve after reconstruction, which was a special examination carried out by a radiologist (Fig. 1), and 14/55 patients were indicated adhesion of the pudendal nerve to surrounding tissues. The indication for SNS was assessed at the pelvic and perineal pain clinic by a medical team, including a neurosurgeon, a neurourologist, a pain physician,

and a physiotherapist. For all recruited PN patients, the indication for SNS should be more cautiously assessed when the pain was debilitating despite the best pharmacological adaptation. Briefly, patients with chronic intractable pelvic and/or urogenital pain were referred to our team's psychologists or psychiatrists to confirm the indication for SNS, and eligible patients were then referred to an implanting physician, or an anesthesiologist-pain physician in a private institution, or a neurosurgeon in the university hospital.

VAS with 0-10 scores was used to assess the pain; the RAND 36-Item Health Survey (SF-36), the Self-rating Depression Scale (SDS), and the Self-rating Anxiety Scale (SAS) were measured at 1 day before operation. Sleep monitors were also carried out at 1 day before surgery and 6 months after surgery. The data were processed and analyzed using Kubios HRV software (Kubios, Kuopio, Finland) and related toolboxes for digital signal processing.

Phase I Test Stimulation

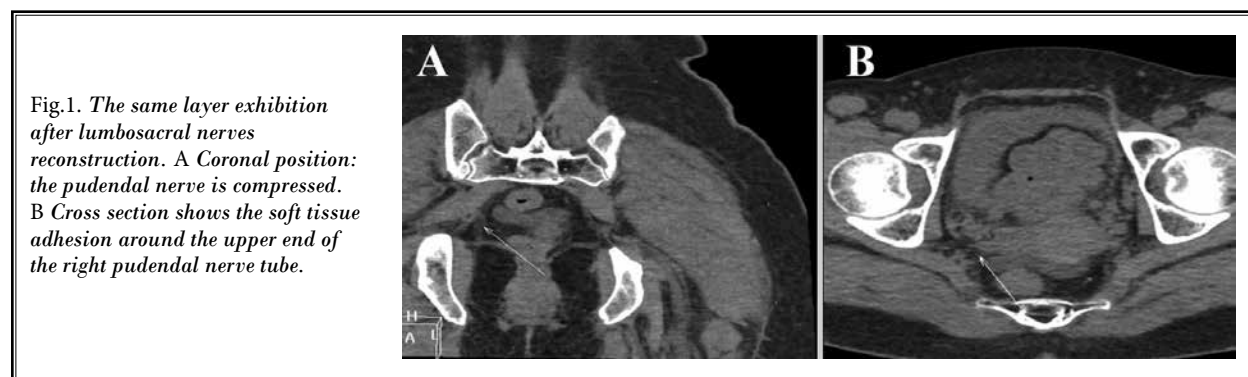
A test stimulation (phase I) was performed for identifying the indication. Spinal cord magnetic resonance imaging and computed tomography (CT) reconstruction were systematically performed to determine the site of puncture. The procedures of the operation, including a CT-guided percutaneous needle puncture and the implantation of stimulation electrodes, were shown in Fig. 2, and the puncture was performed under local anesthesia. The patient's sensory response (rectal traction) and motor response (a contraction of the pelvic floor alone with plantar flexion of the great toe) were tested by a temporary external pulse generator (T901, PINS, Beijing, China) to determine the correct puncture location. The quadripolar self-blocking tined lead (L331, PINS, Beijing, China) is put into the S3 foramen along the lead introducer under the continuous fluoroscopy.

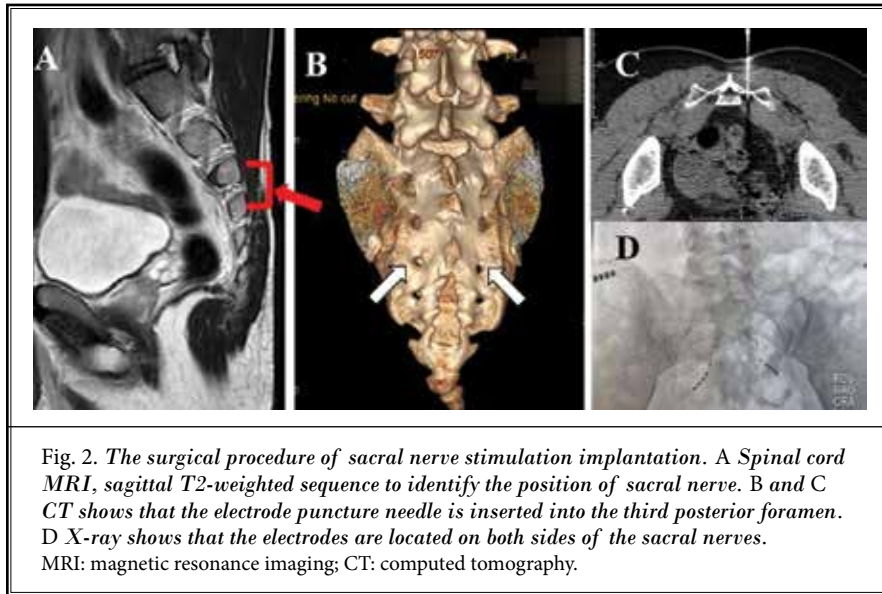
Once the tined lead is positioned, the next step is to tunnel the electrode to a pocket in the buttock and then to connect a temporary external pulse generator (T901) through a lead extension wire for external stimulation during the test period. Bilateral nerve stimulation is per-

Table 1. Descriptive analyses of 33 patients with surgical implantation of permanent stimulator.

Variables	All patients (n = 33)
Age (y), mean ± SD	49.5 ± 11.2
Gender, Women, no (%)	24 (72.7)
Duration of Pain (y), Median, IQR	2.5 (1.3-5.3)
BMI (kg/m ²), mean ± SD	22.7 ± 2.68
A Hot Poker-Like Sensation, no (%)	18 (54.5)
Increased Urinary Urge, no (%)	12 (36.4)
Painful Area, no (%)	
Unilateral Pain	14 (42.2)
Bilateral Pain	19 (57.6)
Pain at the Terminal Branches, no (%)	
Dorsal Clitoris Nerve	15 (45.5)
Perineal Nerve	24 (72.7)
Anal Inferior Nerve	12 (36.4)
Two or More Branches	17 (51.5)
Perineal History, no (%)	
Trauma	7 (21.1)
Surgery	2 (6.1)
Carcinoma	1 (3.0)
Infection	6 (18.2)
Opioid Administration	14 (42.4)
SAS Scale, mean ± SD	61.2 ± 4.6
SDS Scale, mean ± SD	53.0 ± 5.2

Abbreviations: SD: standard deviation; IQR: interquartile range; BMI: body mass index; SAS: Self-rating Anxiety Scale; SDS: Self-rating Depression Scale.





formed for patients with bilateral perineal pain, and the S3 nerve root on the pain side is stimulated for patients with unilateral pain. In addition, the specific response of patients to a subchronic trial of SNS for 2 to 4 weeks was monitored. Patients unresponsive to the test stimulation were tested again or withdrawn, and those with a minimum 50% improvement were candidates for surgical implantation (phase II).

Phase II Permanent Implantation

The patient was readmitted after the test stimulation. The outcomes of test stimulation were assessed one day before the surgery. The subcutaneous stimulator was implanted in patients who were responsive to the test stimulation (a minimum 50% reduction of VAS scores) and informed of implantation after the multidisciplinary meeting. The incision (the pocket as mentioned above) is extended medially under local anesthetic creating a subcutaneous pocket (about 5 cm deep) for IPG placement. The tined lead is connected to the IPG (G131/G132, PINS, Beijing, China) after disconnection of the extension lead and placed within the pocket. Electrodes were removed in patients who had a negative response to the test stimulation or refused the permanent stimulator.

Stimulation Parameters

A positive electrode and a negative electrode are set on the stimulation electrode to be in a bipolar stimulation mode. A positive electrode and 2 negative electrodes set on the stimulation electrode are in a double-

negative stimulation mode. A negative electrode is set on the stimulation electrode, and the stimulator is set to the positive electrode to a unipolar stimulation mode. All patients, who underwent permanent implantation, were in the double-negative stimulation mode.

All patients received constant stimulation. The stimulation intensity ranged from 0.9 V to 2.6 V, among which the intensity of 50% of patients was less than 2.0 V. Stimulation pulse width ranged between 90 μ s and 220 μ s. There were 33%

of patients receiving the stimulation pulse width of less than 100 μ s (narrow pulse width), and the pulse width of other patients were conservative settings (100 μ s~300 μ s). Stimulation frequency ranged between 22 Hz and 40 Hz (high frequency), with a median of 25 Hz, including 25% of patients with a frequency of 22 Hz, 50% of patients with a frequency of 25 Hz, and 25% of patients with a frequency of 40 Hz. No significant modification of stimulation parameters was required during phases I and II, and the long-term follow-up period.

Post-implantation Assessment

Pain assessment was performed the first day after surgery, before discharge (1 week after operation), and 1 month after surgery, then every 6 months. SF-36, SDS, SAS, and sleep monitors were performed at baseline and follow-up (every 6 months). Complications were monitored at each visit.

Statistical Analysis

Statistical analysis was performed with SAS version 9.4 software (Statistical Analysis System Institute, Inc.). Measurement data were expressed as mean \pm standard deviation, and compared using the single factor repeated measures analysis of variance test, and subsequently, multiple comparisons were analyzed by Dunnett's t test. Enumeration data were expressed as percentages, and compared using the chi-square test. $P < 0.05$ considered as statistically significant.

RESULTS

Baseline of Study Population

The 55 PN patients were recruited and subjected to phase I test stimulation for an average of 13 days (6-21 days). Twenty-five percent of patients reported local paresthesia in the perineal area, and other patients also reported paresthesia in the lower limbs, essentially the L4, L5, and S1 dermatomes. The mean reductions in VAS scores were 64% (31%-86%). After multidisciplinary meeting and patient review, 36/55 (65.4%) of patients who had a minimum 50% improvement of pain relief, sleep quality, urination and defecation frequencies, anxiety, and life quality received phase II surgical implantation. One patient lost to follow-up at 6 months because of suicide. One patient was not included in final analysis because of lost contact. Another patient was not included in the final analysis because the subcutaneous stimulator was removed after fractures of the pelvis. Finally, 33 patients, who received the phase II surgical implantation, were included for analysis (Fig. 3).

The analgesic effect of SNS was stable at the last visit compared with that during phase I. Nineteen of thirty-three PN patients experienced bilateral pain. The mean total duration of the pain was 21 months (12-39 months). Six of thirty-three patients had pelvic or genitourinary infections. Eleven had urinary frequency and urgency. The baseline of patients included in our final analysis was shown in Table 1.

Therapeutic Efficacy of SNS

VAS scores were used to evaluate the therapeutic efficacy of SNS in alleviating intractable pelvic and/or urogenital pain in PN patients. There was a favorable decrease in pain severity (VAS scores) from 7.1 ± 1.1 at baseline to 6.1 ± 1.0 on postoperative day 1, and 2.8 ± 0.7 at 1 week, 1.7 ± 0.5 at 1 month, 1.1 ± 0.7 at 6 months, and 1.0 ± 0.6 at 12 months after surgery, respectively (Fig. 4A). In 14 patients (42.4%), opioid administration was needed. The mean opioid consumption (in milligram oral morphine equivalents) decreased after operation, compared to preoperative value (21.4 ± 9.5 vs 3.2 ± 5.8 , $P < 0.05$). The preoperative and 24-hour postoperative opioid consumption among 14 patients was shown in Table 2.

SAS and SDS scores were used to evaluate the therapeutic efficacy of SNS in alleviating anxiety and depression in PN patients, respectively. In particular, SAS and SDS scores were significantly lower at 6 months

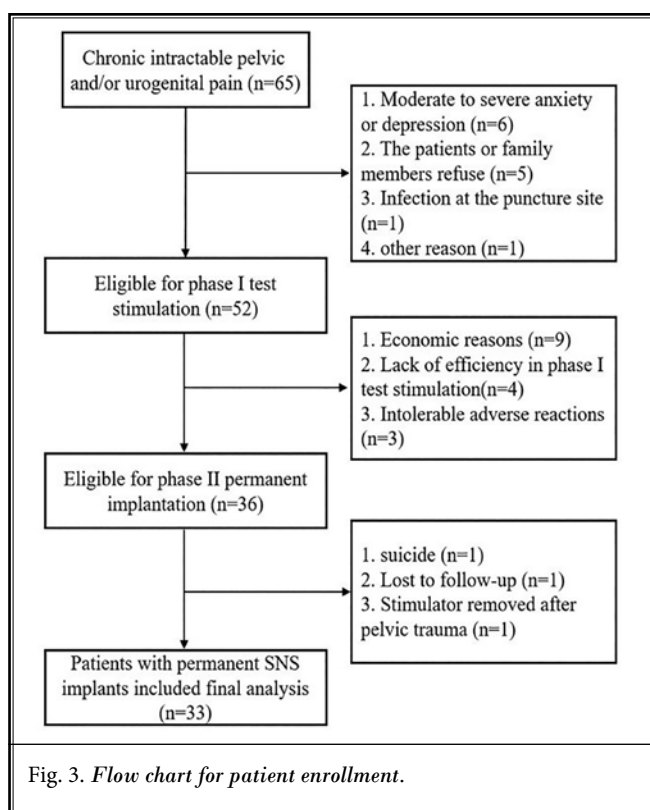


Fig. 3. Flow chart for patient enrollment.

compared with those presurgery after SNS (SAS: 40.1 ± 6.5 vs 53.1 ± 5.3 ; SDS: 44.5 ± 5.6 vs 61.3 ± 4.7 , $P < 0.05$; Figs. 4B and C), while no significant differences were examined between those at 6 and 12 months after SNS (6 months vs 12 months; SAS: 40.1 ± 6.5 vs 40.6 ± 5.8 ; SDS: 44.5 ± 5.6 vs 44.2 ± 5.3 , $P > 0.05$; Figs. 4B and C).

We assessed the quality of life through the SF-36 questionnaire in patients with PN receiving the SNS treatment (Fig. 4D). The mean score of each section of SF-36 after SNS was significantly higher than that at baseline ($P < 0.05$). There was a significant difference in the mean score of each section of SF-36 measured at 6 and 12 months after SNS compared with preoperative levels ($P < 0.05$). However, a significantly higher mean score of SF-36 was only detected in the physical functioning at 12 months after SNS than that at 6 months ($P < 0.05$). The frequency of nocturia was significantly reduced at 6 and 12 months after SNS compared with preoperative levels (6 months vs Pre: 2 (from 1 to 3) vs 3 (from 1 to 7), $P < 0.05$; Fig. 4E).

Sleep Monitoring Indicators

Sleep staging including awake, light, deep, and rapid eye movement (REM) stages from patients be-

fore and 6 months after surgery were monitored (Fig. 5A). Total sleep time and sleep time in each period were significantly prolonged after SNS implantation compared with that before surgery (6 months vs Pre, total sleep time: 5.32 ± 1.49 hours vs 3.66 ± 1.19 hours, deep sleep: 2.52 ± 0.63 hours vs 1.36 ± 0.43 hours, light sleep: 1.78 ± 0.42 hours vs 0.99 ± 0.30 hours, REM sleep: 1.41 ± 0.29 hours vs 0.89 ± 0.27 hours, $P < 0.05$; Fig. 5B). Number of awakenings (6 months vs Pre: 2 (from 1 to 4) vs 4 (from 2 to 8), $P < 0.05$; Fig. 5C) and time spent awake (6 months vs Pre: 0.35 ± 0.14 hours vs 0.55 ± 0.06 hours, $P < 0.05$; Fig. 5D) was also significantly decreased before surgery compared with that at 6 months after surgery. Emergency intensity calculated from heart rate reflecting the stress level of patients before and 6 months after surgery was also obtained. The stress time before surgery was much shorter than that at 6 months after surgery (6 months vs Pre: 3.49 ± 0.47 hours vs 6.71 ± 1.38 hours, $P < 0.05$; Fig. 5E), while the recovery time was significantly prolonged (6 months vs Pre: 1.97 ± 0.39 hours vs 0.55 ± 0.61 hours, $P < 0.05$; Fig. 5E).

Adverse Events

A total of 17 adverse events were noted in the 33 patients with the implant (Table 3). No serious device complications were reported during the follow-up period. Numbness of stimulation area developed in 3 cases, while electrical shock sensation at the implanted neurostimulator site in 7 cases required noninvasive reprogramming. In 4 patients, the pain location changed. Two patients had an initially successful surgical permanent implant, but a wound infection required removal of the IPG. One patient underwent relocation of the IPG due to new pain at the IPG site and also had a dramatic long-term benefit.

Discussion

PN is one of the most disabling symptoms of genital pain. At least 4% of PN patients suffer from chronic pain, which significantly limits daily activities (especially in sitting) and quality of life. So far, PN has become prevalent, and many people sit for a long time every day because of office jobs or journeys (12).

How to effectively alleviate chronic pain in PN patients is challenging, because

the visceral innervation of the pelvic structures share common pathways along the sacral plexus, thus leading to difficulties in identifying the location and cause of pain symptoms (13). Diagnosis of PN often rests on primary diseases-induced symptoms like endometriosis, interstitial cystitis, or irritable bowel syndrome. The full spectrum of associated symptoms is captured by a thorough diagnosis. Therefore, patients with severe voiding complaints, who are well responsive to SNS, may report a concomitant improvement of pain relief. Acknowledging the influence of neuroanatomy on visceral structures contributes to the accurate diagnosis and effective treatment of chronic pelvic pain symptoms. Our study strongly implied that patients with a major complaint of chronic pelvic pain could be

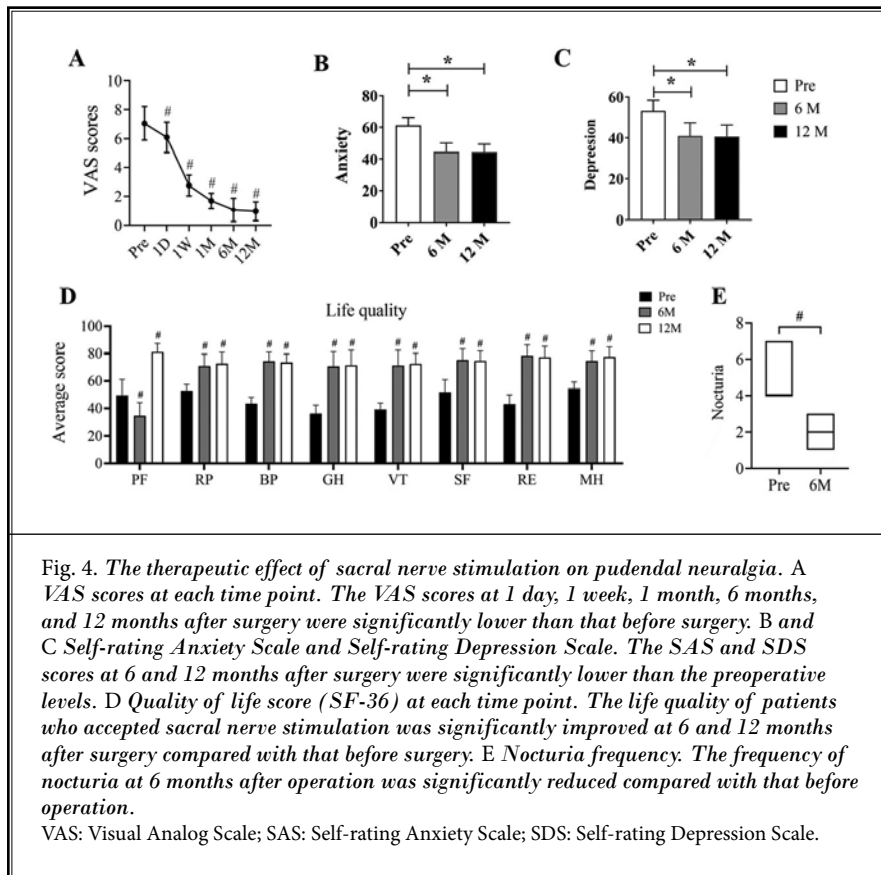
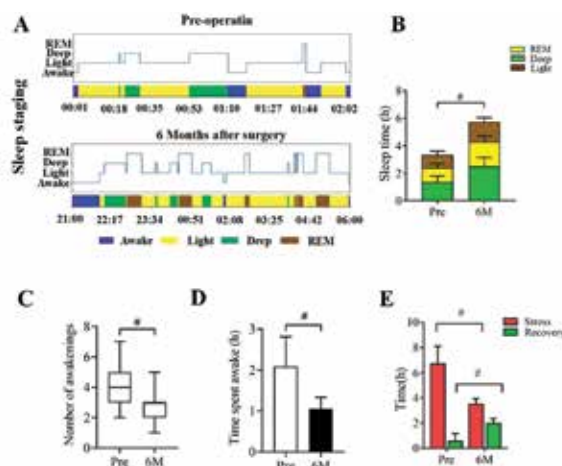


Fig. 5. Sleep improvement after implantation of sacral nerve stimulation. A Representative illustration of sleep staging including awake, light, deep, and REM stages from patients before and 6 months after surgery. B Statistics of sleep staging including awake, light, deep, and REM stages before and 6 months after surgery, indicating that total sleep time and sleep time in each period were significantly prolonged after sacral nerve stimulation implantation compared with that before surgery. C and D Number of awakenings and time spent awake was also significantly decreased before surgery compared with that at 6 months after surgery. E Statistics of stress and recovery time before and 6 months after surgery. The stress time before surgery was much shorter than that at 6 months after surgery, while the recovery time was significantly prolonged. REM: rapid eye movement.



benefited from SNS, and highlighted that pain symptoms may derive from neuromuscular origins (10).

SNS delivers a low amplitude electrical stimulation to persistently stimulate the specific sacral nerves by a subcutaneously implanted stimulator, acting as a neural regulator depriving electrophysiological characteristics of the nerve cells, interfering with abnormal sacral nerve reflex arcs, and mediating effector organ behaviors of sacral innervation. Evidence is accumulating of the treatment of intractable perineal pain with invasive neurostimulations, including stimulations of the peripheral (pudendal) nerve, the sacral root, the dorsal root ganglion, the spinal cord, and the motor cortex. Kim et al (14) have reported the acceptable efficacy of SNS on intractable pelvic pain with cauda equina syndrome (SNS significantly alleviates pain and urinary tract symptoms). Sokal et al (15) have showed a satisfactory short-term effect of SNS in the treatment of 9 women with chronic pelvic pain. Zuidema et al (16) have reported neurostimulation of the S3 nerve root/dorsal root ganglion stimulation via the transforaminal approach in a case with postoperative intractable perineal pain after resection of a vestibular nerve cyst, which relieves pain and improves quality of life. In the present study, VAS scores of 33 patients with intractable perineal pain treated with phase II SNS of stimulator implantation were significantly lower after treatment, and gradually reduced over the follow-up period. The pain was still in relief at 6 months after SNS, suggesting that SNS has a satisfactory short- and medium-term efficacy. Anxiety and depression arising from severe chronic pain can remarkably influence quality of life. Our data showed that SAS and SDS were significantly

Table 2. Daily consumption of morphine in 14 patients.

Patient No	Morphine Equivalent Before Surgery (mg/24 h)	Morphine Equivalent at 6 Months After Surgery (mg/24 h)
1	10	0
2	20	0
3	20	5
4	40	10
5	10	0
6	40	20
7	20	0
8	20	0
9	10	0
10	30	5
11	20	0
12	20	0
13	20	5
14	20	0

Table 3. Adverse events.

Description	No
Numbness of Stimulation Area	3
Electrical Shock Sensation	7
Pain Location Change	4
Impaired Healing of Incision	2
IPG Site Pain	1
Total No	17

Abbreviation: IPG: implantable pulse generator.

reduced after SNS, and the mean score of each section of SF-36 was much higher than baseline level, suggest-

ing that SNS alleviated anxiety and depression, and improved quality of life in PN patients.

Zhang et al (17) have reported that the implantation rate of the stimulator in patients with refractory interstitial cystitis/pelvic floor pain syndrome is 73.1%, and 84.2% of patients have a minimum of 50% improvement in pain symptoms. In our study, 60% of patients were subcutaneously implanted with the stimulator, that is, the ratio of patients with a minimum of 50% improvement after phase I SNS was 63.6%. Previous studies (18) have suggested that the efficacy of SNS on the treatment of perineal pain is uncertain. Dudding et al (18) have reported that 3 of 10 patients with chronic idiopathic anal pain have at least 50% reduction in VAS scores after SNS, and only 1 of 10 show a good control of pain during the 5-year follow-up. We consider that the nociceptive input of intractable perineal pain exists at multiple levels of the nervous system, and as a result, multilevel targets of SNS exist. In addition, the anatomic structures of the perineal area are complicated, especially in the branches of the sacral nerve. Inflammatory edema in local neuropathy can also influence the efficacy of SNS. In addition, reprogramming is an essential tool in optimizing SNS therapy. Up until now, SNS has most often been applied as a rectangular signal, with a stimulation frequency of 14 Hz and a pulse width of 210 μ s, also referred to as standard (conservative) stimulation parameters (2). Whether the change of SNS stimulation parameters may result in an improved outcome has been controversial. Studies (3,4) of spinal cord stimulation in treatment of neuro-

modulation have shown that long-term efficacy can be improved with the use of new stimulation parameters. However, there are no studies reporting the role of changes in SNS stimulation parameters in perineal pain. We found that high frequency (> 20 Hz) may be better for patients with PN to relieve their pain. It is hoped that this finding will provide pointers for future research focusing on SNS stimulation parameters in perineal pain.

As the pudendal nerve is derived from the S2, S3, and S4 nerve roots, the S3 nerve root and the conus medullaris are 2 promising stimulation targets. Retrograde stimulation of the S3 nerve root is a relatively difficult clinical procedure with a high failure rate (9,19). In our experiences, the electrode movement of S3 nerve root stimulation is the main challenge that influences the stable long-term efficacy. Stimulation of the conus medullaris has been previously reported in 2 patients (20,21). In clinical practice, the spinal cord stimulation is conventionally used in the treatment of refractory neuropathic pain of the lower limbs.

CONCLUSIONS

Collectively, SNS is a simple, conservative technique that effectively prolongs the sitting time and enhances the estimated percent improvement in PN patients with chronic intractable pelvic and/or urogenital pain after failure of surgical decompression of the pudendal nerve. The efficacy of SNS appears to be stable over time. Our findings should be further validated in large-scale prospective studies.

REFERENCES

1. Robert R, Prat-Pradal D, Labat JJ, et al. Anatomic basis of chronic perineal pain: Role of the pudendal nerve. *Surg Radiol Anat* 1998; 20:93-98.
2. Spinosa JP, de Bisschop E, Laurencon J, et al. [Sacral staged reflexes to localize the pudendal compression: An anatomical validation of the concept]. *Rev Med Suisse* 2006; 2:2416-2418, 2420-2421.
3. Rhame EE, Levey KA, Gharibo CG. Successful treatment of refractory pudendal neuralgia with pulsed radiofrequency. *Pain Physician* 2009; 12:633-638.
4. Buffenoir K, Rioult B, Hamel O, et al. Spinal cord stimulation of the conus medullaris for refractory pudendal neuralgia: A prospective study of 27 consecutive cases. *Neurol Urodyn* 2015; 34:177-182.
5. Pereira A, Perez-Medina T, Rodriguez-Tapia A, et al. Chronic perineal pain: Analyses of prognostic factors in pudendal neuralgia. *Clin J Pain* 2014; 30:577-582.
6. Cok OY, Eker HE, Cok T, et al. Transsacral S2-S4 nerve block for vaginal pain due to pudendal neuralgia. *J Minim Invasive Gynecol* 2011; 18:401-404.
7. Robert R, Labat JJ, Bensignor M, et al. Decompression and transposition of the pudendal nerve in pudendal neuralgia: A randomized controlled trial and long-term evaluation. *Eur Urol* 2005; 47:403-408.
8. Matzel KE, Chartier-Kastler E, Knowles CH, et al. Sacral neuromodulation: Standardized electrode placement technique. *Neuromodulation* 2017; 20:816-824.
9. Shaker H, Hassouna MM. Sacral root neuromodulation in the treatment of various voiding and storage problems. *Int Urogynecol J Pelvic Floor Dysfunct* 1999; 10:336-343.
10. Siegel S, Paszkiewicz E, Kirkpatrick C, et al. Sacral nerve stimulation in patients with chronic intractable pelvic pain. *J Urol* 2001; 166:1742-1745.
11. Labat JJ, Riant T, Robert R, et al. Diagnostic criteria for pudendal neuralgia by pudendal nerve entrapment (Nantes criteria). *Neurol Urodyn* 2008; 27:306-310.
12. de Tayrac R, Haylen BT, Deffieux X, et al. [French translation of "An International Urogynecological Association (IUGA)/

- International Continenence Society (ICS) joint report on the terminology for female pelvic floor dysfunction” published in *Int Urogynecol J* 2010; 21:5-26]. *Prog Urol* 2016; 26:197-225.
13. Ryder RM. Chronic pelvic pain. *Am Fam Physician* 1996; 54: 2225-2232, 2237.
 14. Kim JH, Hong JC, Kim MS, et al. Sacral nerve stimulation for treatment of intractable pain associated with cauda equina syndrome. *J Korean Neurosurg Soc* 2010; 47:473-476.
 15. Sokal P, Zielinski P, Harat M. Sacral roots stimulation in chronic pelvic pain. *Neurol Neurochir Pol* 2015; 49:307-312.
 16. Zuidema X, Breel J, Wille F. S3 dorsal root ganglion/nerve root stimulation for refractory postsurgical perineal pain: Technical aspects of anchorless sacral transforaminal lead placement. *Case Rep Neurol Med* 2016; 2016:8926578.
 17. Zhang P, Zhang JZ, Wu LY, Niu HQ, Yang YB, Zhang XD. Short-Term outcome of sacral neuromodulation on refractory interstitial cystitis/pelvic pain syndrome. *Zhong Hua Yi Xue Za Zhi* 2016; 21:573-576.
 18. Dudding TC, Thomas GP, Hollingshead JR, et al. Sacral nerve stimulation: An effective treatment for chronic functional anal pain? *Colorectal Dis* 2013; 15:1140-1144.
 19. Antolak SJ Jr, Antolak CM. Therapeutic pudendal nerve blocks using corticosteroids cure pelvic pain after failure of sacral neuromodulation. *Pain Med* 2009; 10:186-189.
 20. Reichart R, Kuhn SA, Vogel I, et al. [Spinal cord stimulation at the level of the conus medullaris: Treatment option for therapy-resistant postoperative neuralgia of the pudendal nerve]. *Schmerz* 2009; 23:640-644.
 21. Rigoard P, Delmotte A, Moles A, et al. Successful treatment of pudendal neuralgia with tricolumn spinal cord stimulation: Case report. *Neurosurgery* 2012; 71:E757-E762; discussion E763.

