EXPERIENCE OF COMBINED PUDENDAL AND SACRAL ELECTROSTIMULATION IN A PATIENT WITH CHRONIC PELVIC PAIN

© A.A. Polushkin 1, E.D. Isagulyan 1, A.A. Tomskiy 1, R.V. Salyukov 2

1 N.N. Burdenko National Medical Research Center of Neurosurgery, Moscow, Russia;
2 RUDN University of the Ministry of Science and Higher Education of the Russian Federation, Moscow, Russia


Treatment of patients with chronic pelvic pain is one of the most difficult tasks of modern medicine. Recently, surgical neuromodulation has been increasingly used to treat chronic pain syndrome. The positive experience of chronic sacral stimulation usage in patients with pelvic organ dysfunction accompanied by pain syndrome determines the appropriateness of further search for effective methods of chronic pelvic pain treatment, for example, its combination with stimulation of peripheral nerves. The article presents a clinical observation of a patient suffering from chronic pelvic pain for a long time, who underwent chronic electrical stimulation of the sacral roots in combination with sacral nerve stimulation. After the first year of treatment, a decrease in pain intensity on the visual analog scale from 8 to 3 points, depression on the Zips from 14 to 10 points and a decrease in the indicator on the anxiety scale from 14 to 11 points was observed. The patient’s quality of life was improved, pain attacks were decreased, daily motor activity was increased, self-service along with social communication indicators were improved and the need for medicines was decreased.

Keywords: chronic pelvic pain; neuromodulation; sacral neuromodulation; pudendal nerve stimulation.

CASE REPORTS / Клинические наблюдения
According to the definition of the European Association of Urology, chronic pelvic pain (CPP) is a constant or persistent pain that is perceived in structures related to the pelvic organs in men and women. Pelvic pain typically develops with a certain disease of the internal organs (oncological, inflammatory, and traumatic) and chronic pelvic pain syndrome (CPPS), which at the time of consultation does not show a clear connection between the pain and any morphological change in the pelvic organs or the musculoskeletal system forming it [1, 2]. CPPS patients do not reveal any obvious local pathological changes that may explain the pain.

According to the International Association for the Study of Pain, the prevalence of CPP is 6%–7% [3]. Chronic pelvic pain is common in women of reproductive age. Women with complaints of CPP symptoms account for 15%–20% of all patients in gynecological consultations; they represent up to 10% of all cases of women applying to general practitioners [4].

An increase in the number of CPP patients with unsatisfactory results of conservative treatment has led to the search for new approaches, methods, and ways to solve this problem. One of the most effective and promising methods of CPPS treatment is neuromodulation, which involves electrical stimulation of sacral roots. With an increase in the use of neurostimulation to improve the functions of the pelvic organs, its application in CPP treatment has expanded with a positive effect. Good results in CPPS control through spinal cord stimulation have also been noted. Over the past decade, many works have been published on the stimulation of the genitals and other nerves involved in innervation of the small pelvis organs and structures in CPPS treatment. Several studies have also been published, which compare the efficacy of various types of neurostimulation in CPPS treatment. However, long-term results are not as good as expected, according to a follow-up of the test or early postoperative periods. In some cases, these results are due to progression of concomitant psychological problems that often accompany CPPS or have perioperative complications in other cases. Deterioration in the prospective follow-up history is often explained by the "habituation phenomenon" and is often the reason for the unclear effect depletion. In fact, this condition is similar to the root cause of CPPS, which remains unclear in almost a third of cases. In this regard, the combination of various methods of neuromodulation (neurostimulation, and the combination of various methods of neuromodulation with intrathecal drug delivery) reveals new prospects in the treatment of such patients. We present an example of the combined use of the aforementioned methods, including sacral and pudendal electrical stimulation.

CLINICAL CASE

Patient N., 54 years old, was admitted with complaints of constant burning pain in the urethra, most pronounced after urination. The anamnesis reveals that the patient has been suffering from pain for 8 years. In adolescence, she experienced acute cystitis several times. The gynecological history shows two pregnancies and two births. The first birth was at age 23, with a large fetus with an episiotomy. During her visit to the gynecologist, a stressful form of urinary incontinence was detected and confirmed by urodynamic examination. Sling urethropexy was performed with a positive clinical effect, and continence was restored. No changes were reported in the intensity and nature of pain after surgery.

For the pain syndrome, the patient was repeatedly examined by an urologist and a gynecologist. Diseases that could cause severe urogenital pain were not detected. For five years, the patient was observed by a neurologist. During that period, various generally accepted international treatment regimens for neuropathic pain have been tried, based primarily on different combinations of anticonvulsants, antidepressants, and local anesthetics. The latter were used in the form of applications and blocks of trigger points and the pudendal nerve area. A psychotherapist prescribed clomipramine, which had a temporary positive effect. At the time of hospitalization, the patient was taking clomipramine (25 mg/day) and alimemazine tartrate (5 mg/day). To relieve severe pain, she had to resort to oral and sometimes parenteral administration of Tramadol (50–150 mg per day).

The pain was constant, varied in intensity from 3 to 8 points during the day based on a visual analogue scale (VAS), and was 6 points most of the day. The pain was localized mainly in the urethra and perineum, more to the right, as well as in the area of the clitoris and external labia, also mainly on the right. Lumbago occurred from the perineum to the rectal region up to 3 times a day with constant pain. Neither background nor paroxysmal pain was directly related to a specific position. However, with deep palpation of the pudendal nerve exit site (in the area of the sciatic spine), a distinct soreness was noted on
the right, which spread to the perianal region, partially reproducing the patient’s typical pain. In addition, the pudendal nerve blockade at this point led to a short-term (several hours) but distinct regression of the pain syndrome. Bulbocavernous and anal reflexes were preserved. Pelvic functions were controlled, and the anal sphincter had sufficient tone. Urination was painless and partly difficult, especially the initiation process. After urination, as a rule, an increase in burning occurred in the area of the urethra and labia with irradiation to the perianal region. Cotton-swab test showed a strong positive result.

The neuropathic nature of the pain syndrome was confirmed using standard international scales [5], namely, 4 positive answers in Douleur Neuropatique 4 questions, 10 points in Pain Detect, and 17 points in Leeds Assessment of Neuropathic Symptoms and Signs.

Psychological status was assessed on the scale of anxiety and depression in Hospital Anxiety and Depression Scale. The testing results revealed a clinically pronounced level of anxiety and depression. The effect of pain on various parameters of quality of life (QOL) was determined using a modified brief pain inventory questionnaire, Pain Quality of Life Card (PQLC) [5–7]. This questionnaire revealed a significant decrease in the QOL due to severe pain syndrome.

Given the neuropathic nature of the pain and its refractivity to conservative treatment, we established indications for neurostimulation. Aspects of localization and a wide irradiation of pain within the small pelvis, as well as a combination of pain syndrome with urination difficulty, indicated the suitability of sacral stimulation. However, in this patient, a somewhat unusual pattern of pain syndrome was determined by the involvement of the pudendal nerve directly in its pathogenesis, as indicated by local soreness and positive results of the blockade. Based on these findings, combined stimulation was performed on the sacral roots and pudendal nerve from the side where the pain prevailed, that is, to the right.

In March 2017, two electrodes were immediately implanted into the patient in the third sacral foramen on the right and the pudendal nerve on the right. The intervention was conducted in a specialized radiological operating room under local anesthesia with intravenous potentiation with Propofol. Before surgery with the patient in prone position with rollers placed under the pelvic bones and ankle joints, the third sacral foramen was marked and the projection of the pudendal nerve trunk in the area of the sciatic spine on the right was designated.

The third sacral foramen (S3) was marked according to the generally accepted method under radiological control [8]. As a rule, it is projected onto the skin at the intersection of the vertical line drawn through the medial edges of the sacral foramen with the horizontal line drawn along the lower edge of the sacroiliac joint (Fig. 1).

The pudendal nerve trunk is short, and it enters the pelvic area from the sub-piriform space and then passes through the pudendal canal, at the exit of which it branches. Thus, the main place where the nerve trunk can be influenced corresponds to its location next to the sciatic spine of the sciatic bone. The marking for electrode implantation in this area was also performed under X-ray control according to the STAR method (spine, tuberosity, acetabulum, and analrim) [9]. Initially, on the implantation side of the electrode, the middle of the acetabulum was determined, which corresponds to point A. A horizontal line was drawn on the skin through point A with a marker. Then, the point T corresponding to the middle of the lower edge of the ischial tuberosity was determined. A vertical line was drawn through the point T perpendicular to the first one. The intersection of these lines (point S) corresponds to the sciatic spine, which is also visible in the oblique image (with an approximately 15–30° angle of inclination), because in a strictly direct projection it is overlapped with the hip joint or branch of the sciatic bone. From point S, a line was drawn to the outer edge of the anal sphincter (point R). After the points T and R were connected, the resulting segment was divided in half.

Fig. 1. The X-ray of the pelvic bones in a direct projection indicating anatomical landmarks and conditional lines drawn through them to determine the hole of the S3, the arrow indicates the edge of the iliosacral joint, the circle on the S3 indicates the hole (left). On the right the a projection of the third sacral hole is visualized

Рис. 1. Рентгенограмма костей таза в прямой проекции с указанием анатомических ориентиров и проведенных через них условных линий для определения отверстия S3, стрелкой указан край ильиосакрального сочленения, кругом S3 обозначено отверстие (слева). Справа на снимке металлическим инструментом указаны проекции третьего сакрального отверстия
The middle of this segment served as the point for the needle insertion (yellow circle) and the apex of the triangle (point S) was the target for advancing the needle with the electrode (red circle) (Fig. 2).

At stage 1, an electrode was implanted in root S3. After a short skin incision (up to 2.0 cm) before aponeurosis, pockets were formed by stratification of subcutaneous fatty tissue from it to lay the electrode loops and site of its connection with a temporary extension cable. To install the electrode, we used a standard Tuohy needle, which was directed almost perpendicular to the foramen S3 to the bone. Thereafter, at a 60° angle to the surface and about the same in relation to the midline, the needle was further moved deeper into the tissue until the feeling of loss of resistance and the tip of the needle appeared at the lower edge of the sacrum in a lateral projection (Fig. 3). After removal of the stylette, an electrode was drawn along the needle lumen. When positioned correctly, it should have a craniocaudal direction in lateral projection and a mediolateral direction in a direct radiograph (see Fig. 3).

In addition to radiological support, neurophysiological control, particularly intraoperative electrical stimulation, was used to confirm the accuracy of the electrode location. With an adequate electrode position, stimulation with amplitude of less than 2.0 mA causes plantar flexion of the great toe and reduction in the anal sphincter.

At stage 2, an electrode was implanted in the region of the trunk of the pudendal nerve. To perform this, we made a skin incision up to 2 cm long in the area of the starting point determined using the STAR technique; a subcutaneous pocket was also formed on either side of the incision. The Tuohy needle was set at an angle of 50–60° to the buttock skin, tilted to the medial side at 15–20° and, under X-ray control, was directed to the sciatic spine (point S), when reaching with the needle tip was advanced by another 1.0 cm. The electrode was inserted through the lumen of the needle. The correctness of its position was also controlled by X-ray and intraoperative stimulation (contraction of the anal sphincter with amplitude of less than 2.5 mA).

To prevent displacement of electrodes in tissues, we fixed them with specially adapted silicone “anchor” devices (Fig. 4). Thus, two cylindrical eight-contact electrodes were implanted into the patient, which were connected to temporary extension cords, with the distal ends brought out through the counterincision outward in the lumbar region on the right. The electrode excess was folded as rings and, together with the electrode joints with temporary extension cords, were placed in the subcutaneous pockets. The wounds were sutured tightly. For test stimulation, the ends of the connectors brought out through counterincision were connected using a special cable with a screener, which was a temporary external stimulator.

A day after the surgery, X-ray control of the position of the electrodes was conducted (Fig. 5).

The parameters of electrical stimulation were set for each electrode individually, namely, frequency of 110 Hz, and pulse width of 210 and 240 ms with possibility of changing the amplitude from 2.0 to 10.0 mA. In the process of selecting stimulation programs, we tried to achieve full coverage of the pain zone with a sensation of a uniform, pleasant vibration. The test stimulation effectiveness was evaluated using a special diary on which the patient indicated the pain syndrome intensity on the VAS scale before, during, and after electrical stimulation. During the
test period, isolated stimulation of the third sacral root was initially performed. During this stimulation, the patient did not notice a complete overlap of the pain area, and the urge to urinate was increased. Analysis of the program showed that at a pulse width of 300 ms, the patient felt a more pleasant vibration compared with a width of 210 ms. With isolated stimulation of the pudendal nerve, the pain area also was not overlapped completely, and the patient did not feel the difference in pulse width for a pleasant vibration. However, to achieve the analgesic effect, the stimulus amplitude was required to be above 7 mA. With simultaneous stimulation of the sacral root and pudendal nerve, the pain area was overlapped completely. In this case, the vibration sensation was stronger with a pulse width of 240 ms and stimulus amplitude of 5 mA. Thus, combined stimulation was most effective. The test period was 12 days. During test stimulation, the patient noted a 50% decrease in pain on the VAS scale (from 8 to 4 points), which indicates an implantation of a system for chronic neurostimulation.

On April 11, 2017, under general anesthesia a subcutaneous generator of the S3 root and right side of pudendal nerve chronic stimulation system was implanted. The surgery was performed according to standard methods. The junction of the electrodes with temporary extension cords was initially allocated; the latter was disconnected and removed. Then, a subcutaneous pocket for a neurostimulator was formed in the upper part of the left gluteal region, and the proximal ends of the electrodes without temporary cables were tunneled subcutaneously into this pocket. The latter were inserted into a pulse generator that was fixed in the pocket with interrupted sutures. All wounds were sutured tightly with subcuticular sutures.

The stimulation parameters determined in the test period were set for the patient. After the implantation of the pulse generator, considering the data obtained in the test period, simultaneous stimulation was possible in which stimulation was not only “parallel” but “cross” stimulation. Principle of this method is as follow. With parallel stimulation, paresthesia can be achieved separately in the area corresponding to the innervation of a particular nerve or root. With cross stimulation, one of the electrodes can be used as a cathode and the other as an anode. In this case, the electric field becomes much wider and the paresthesia zone increases, while the paresthesia itself becomes much milder and more pleasant. In this case, we installed cathodes on the electrode in the pudendal nerve area and anodes on the sacral electrode. With such a program, the patient noted not only a complete coverage of the pain area with pleasant paresthesia, but also a significant reduction in pain by more than 50%. The median pain regression amounted to 65%. We did not register any surgical and neurological complications in either early or late postoperative period in this patient.

Analysis of the treatment efficiency was performed 6 and 12 months after surgery. The stable analgesic effect of stimulation, predominantly in the "cross" mode, significantly reduced the severity of the pain syndrome and its effect on various parameters of the QOL. Given the stable effect, further adjustment of stimulation programs has not been performed yet.

When comparing the results of the questionnaire survey before the use of neurostimulation and after 12 months in the course of stimulation, we obtained...
the following results. The VAS pain intensity indicator before constant stimulation was 8 points, and 3 points after a year (pain was reduced by approximately 65%). The severity of depression before surgery was 14 points according to HADS, and 10 points after a year. The indicator on the anxiety scale was also initially 14 points, and 11 points after a year.

When analyzing the effect of neurostimulation on the patient’s QOL, we observed an improvement on the PQLC scale in many areas. Thus, the average value of the most severe background pain on the PQLC scale before stimulation was 8 points, and 5 points during the stimulation after a year. The most severe pain attack was 10 points, and 7 points during the stimulation after a year. The severity of the weakest background pain before surgery was 7 points, and 3 points during the stimulation after a year. The weakest attack of pain before surgery was 5 points, and 3 points during the stimulation after a year. The frequency of seizures was 9 points, and 4 points during the stimulation after a year. The severity of the weakest pain attack was 10 points, and 7 points during the stimulation after a year. The most severe scale before stimulation was 8 points, and 5 points during the stimulation after a year. The average intensity of background pain was 6 points, and 4 points during the stimulation after a year. The average intensity of a pain attack was 7 points, and 3 points during the stimulation after a year. The need for medication before implantation was 12 points, and 6 points during the stimulation after a year. The effect of pain on well-being was 5 points, and 2 points during the stimulation after a year. The effect of pain on mood was 10 points, and 7 points during the stimulation after a year. The effect of pain on daily motor activity was 8 points, and 4 points during the stimulation after a year. The effect of pain on passive rest was 8 points, and 5 points during the stimulation after a year. The effect of pain on self-care was 7 points, and 3 points during the stimulation after a year. The effect of pain on relationships with other people was 2 points, and 2 points during the stimulation after a year. The effect of pain on sleep was 5 points, and 4 points during the stimulation after a year. The effect of pain on sexual activity was 10 points, and 8 points during the stimulation after a year.

Thus, when the treatment was combined with neurostimulation, a persistent good analgesic effect (during the year) was achieved. The most significant effect of treatment on QOL was a decrease in the number of pain attacks, an increase in daily locomotor activity, an improvement in self-care and social communication, and a decrease in the need for medication.

**DISCUSSION OF RESULTS**

Experience has confirmed the efficiency of neuromodulation in CPP treatment [10–14]. For instance, Martellucci et al. cited data from a prospective multicenter study of the method in the treatment of 27 CPPS patients. A pronounced decrease in pain was achieved in 16 (59%) patients when neurostimulation was used consistently during the follow-up year [15]. Peters and Konstandt demonstrated the effectiveness of sacral neuromodulation in the treatment of urinary bladder pain in 21 patients. Decrease in pain severity within 15 months of follow-up was achieved in 95% of the patients. The authors noted that 18 patients managed to reduce the consumption of painkillers by 36% [16].

Today, implantation of an electrode to the S3 root can be considered a classical surgical technique for sacral neuromodulation. Thus, attempts to implant electrodes to other sacral roots are also interesting. For example, Siegel et al. [17] stimulated not only S3 roots but also S4 spinal nerves. A study by Sherman et al. [18] suggested that stimulation of the S3 root is inferior to the effect on the pudendal nerve because it contains fibers of the S2–S4 roots.

Fan et al. [19] shared the unique experience of successful implantation of electrodes on the pudendal nerve in CPP patients for whom sacral neuromodulation was not effective. The authors showed that pudendal nerve stimulation can be effective in patients with unsuccessful sacral stimulation.

In 2004, Peters et al. [20] published the results of a randomized, blind study comparing sacral and pudendal stimulation with urinary dysfunction. The study included 30 patients who received a sacral and pudendal electrode implanted simultaneously. The patients did not know which electrode was programmed for stimulation. At the end of the study, 79.2% of patients noted an improvement in the pelvic organ function during stimulation of the pudendal nerve, while in 20.8%, improvement was noted with stimulation of the S3 root. In addition to improvement of the pelvic organ functions, the patients noted a more pronounced decrease in pain during stimulation of the pudendal nerve than during electrical stimulation of the sacral root.

In the clinical case described, we used a combined neurostimulation technique in which one electrode is placed in the S3 root region and the other is placed in the pudendal nerve region on the right. Both methods are low-traumatic. During chronic stimulation, the patient was able to significantly reduce the severity of the pain syndrome by more than 50% while main-
taining a stable analgesic effect throughout the year, as well as improve the psychological status, improve the QOL, and reduce the amount of drugs taken.

Despite the many years of successful use of surgical neuromodulation, unfortunately, these methods have not yet become standard in the treatment of patients with CPP syndrome. In most cases, neuromodulation techniques are recommended for use only if standard treatment methods are ineffective. However, further study of the possibilities of neuromodulation as the main treatment method for CPP, such as refractory to conservative treatment, is necessary. We also need to determine precisely not only the criteria but also the temporal parameters of refractoriness, which, as a rule, should not exceed six months because each subsequent month severely worsens the prognosis and reduces the possibilities of neurostimulation techniques.

In the clinical case we presented, the CPPS diagnosis was established based on an interdisciplinary approach that seemed to be the most correct, especially because this diagnosis was conducted by exclusion.

**CONCLUSION**

The clinical case described was the first Russian experience of combined stimulation of the sacral root S3 and pudendal nerve. This case reflected the general vector of development of contemporary functional neurosurgery in CPP treatment. The positive treatment result indicates the prospect of further study on the possibilities of peripheral neurostimulation in CPP treatment.

**REFERENCES**


