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Sacral roots stimulation in chronic pelvic pain

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ABSTRACT

Introduction: Chronic pelvic pain is a syndrome of chronic non-malignant pain of multifactorial pathophysiology. Perineal, anal and coccygeal pain can be a form of failed-back surgery syndrome or complex regional pain syndrome. Apart from conservative treatment interventional methods are useful in this condition as neurolytic blocks or non-destructive neuromodulation procedures. Peripheral nerve, spinal cord stimulation or sacral stimulation can be applied.

Aim: We describe a minimally invasive method of sacral roots stimulation with percutaneous electrodes implanted through the sacral hiatus in the treatment of chronic pelvic pain.

Materials and methods: We evaluated a series of nine female patients with pelvic pain treated with sacral roots stimulation in regard of efficacy and complications of this method.

Results: Short-term results in all patients were satisfactory with statistically significant improvement (median VAS = 9 before surgery) (median VAS = 2 after implantation, $p = 0.001$), (median VAS = 3 after 6 months, $p = 0.043$). The long-term follow-up revealed less satisfactory result (median VAS = 6 after 12 months). High incidence of complications was noted: mainly infection in 3/9 patients.

Conclusion: Sacral roots stimulation is a non-destructive and minimally invasive neuromodulation method in the treatment of chronic pelvic pain. It can be effective even in the long-term observation but special care is advised to secure aseptic conditions in the implantation and to prevent the infection which leads to removal of the stimulating system.

1. Introduction

Chronic pelvic pain (CPP) is a syndrome of chronic non-malignant pain of multifactorial pathophysiology occurring in both sexes. The etiology and pathogenesis of this condition is poorly understood and can be associated with disturbances in skeletal, genitourinary, gastrointestinal systems. It has severe effects on quality of life. The type of pain in CPP is usually complex: visceral pain originating from organs of minor pelvis, somatic when it comes from bones, joints or muscles of this region and neuropathic one when the injury of peripheral or central nervous system occurs. Coccycodynia is the occurrence of pain in the coccyx region of unclear origin. In most cases a traumatic etiology is present [2]. The management of CPP is multidisciplinary and involves several specialists. CPP

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can be treated conservatively with pharmacotherapy using antibiotics and non-steroid anti-inflammatory drugs (NSAIDs) in inflammatory conditions, antiepileptic drugs as gabapentine, antidepressants as amitriptyline or muscle relaxants in neuromuscular disorders. In the acute phase the first choice of treatment are NSAIDs. The physiotherapy and psychotherapy are important adjuvant interventions. When conservative treatment is insufficient more aggressive methods can be applied. Neurolytic blocks with local anesthetics and steroids or radiofrequency lesions can be recommended [3,4]. Particularly neurolytic blocks of the ganglion of Walther are efficacious with long-term relief and the low rate of complications [5]. In coccycodynia coccygectomy is not recommended because of long-term moderate effects and increased risk of major complications [2]. The CPP with predominance of neuropathic pain can be the part of failed-back-surgery syndrome (FBSS) or complex regional pain syndrome (CRPS) which are indications for neuromodulation [3]. CPP is supposed to be a form of CRPS and can fill criteria of this syndrome [6,7]. Neuromodulation procedures are not destructive but reversible. Spinal cord stimulation (SCS) is the most popular and widespread type from neuromodulation techniques. The reports of the effects of spinal cord or spinal conus stimulation in CPP are not conclusive. The optimal localization of electrode in SCS is not defined and is to be considered and discussed. An interesting and minimally invasive method of treatment of CPP is peripheral nerve stimulation (PNS): pudendal, illo-inquinal, genito-femoral or tibial nerve stimulation [8]. Another option is a targeted stimulation (TS) in the field of the highest intensity of pain [4]. A sacral neuromodulation (SN) is an electrical stimulation of sacral nerves which has to modulate the function of neural reflexes and pain afferent tracts. SN is an established therapy for lower urinary dysfunction approved by FDA for urgency-frequency and urinary incontinence. SN has been shown to have benefits in painful bladder syndrome/interstitial cystitis, urinary urge incontinence, urinary nonobstructive retention, urinary urgency as well as frequency and stool incontinence [9–11]. The success rate after permanent implantation ranges from 66% to 77% in patients responding to test stimulation [12]. SN is supposed to show better benefits than standard medical therapy [13]. The reduction of pain syndromes is observed in both SN and PNS with better result in SN [14,15]. In SN transforaminal implantation unilateral or bilateral is applied. Sacral roots stimulation (SRS) is a neurostimulation of sacral roots S2, S3, S4. SRS can be performed with anterograde, retrograde percutaneously implanted electrodes.

The mechanism of the action of SN or SRS is based on Melzack and Wall' gate theory [16]. It has to block afferent pain transmission, activate descending inhibitory pathways, effect on sympathetic system, modulate neuromediator activity in dorsal horns.

2. Aim

We describe a minimally invasive, neuromodulative method of the stimulation of the sacral roots with percutaneous electrodes implanted through sacral hiatus in the treatment of CPP based on our experience.

3. Materials and methods

We evaluated the effects of SRS in the treatment of CPP in nine female patients hospitalized in the Department of Neurosurgery of Military Research Hospital in Bydgoszcz Poland in years 2008–2013. Median age was 57 years, ranging from 41 to 77 years. Median duration of pain was 6 (range from 5 to 17). Follow-up was from 1 to 48 months.

All patients gave the informed consent on surgical treatment defining risks, potential benefits and on evaluation of the effects of procedures, which were performed by personal interview or clinical examination.

We assessed the intensity of pain before the treatment, after the surgery, after 6 months and after 1 year in visual analog scale (VAS). Depending on localization, etiology and causative factor CPP in nine patients was divided on FBSS (Nr = 4) and idiopathic CRPS (Nr = 5). Cases 1, 2 were treated with SCS at the beginning with electrode localized epidurally on the level of spinal conus, e.g.: Th12-L1. Surgery was performed under general anesthesia and electrodes were implanted through flavectomy or partial laminectomy on appropriate level under fluoroscopic control. Due to poor results of SCS we performed SRS in these patients. All nine patients underwent the implantation of sacral percutaneous electrodes (two or one; quadripolar or octopolar) through the

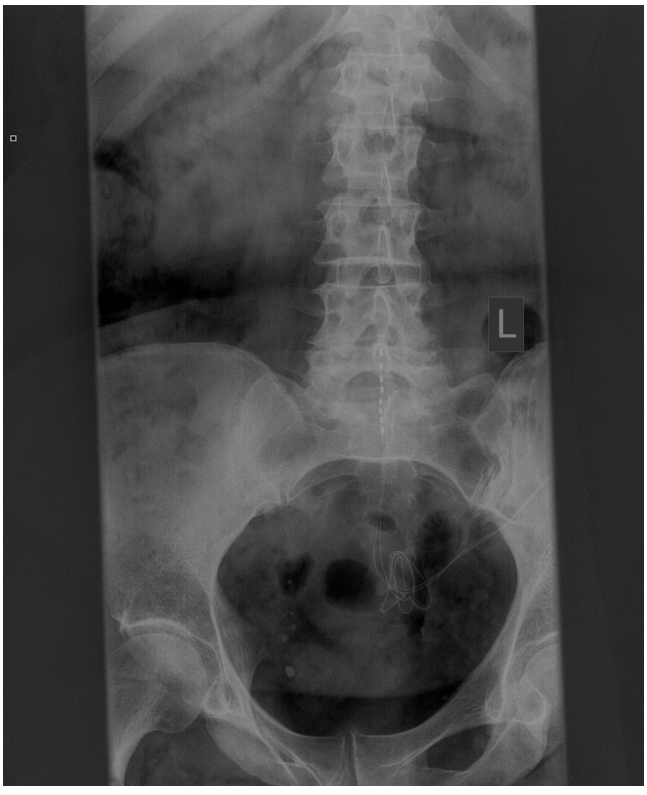

Fig. 1 – A 56-year-old female patient with perianal, burning, idiopathic pain lasting 6 years, NRS = 9 treated with tramadol and gabapentin. Two quadripolar electrodes implanted into the sacral canal. Result 90% reduction of pain. Due to infection after 1 month – removal of the system.

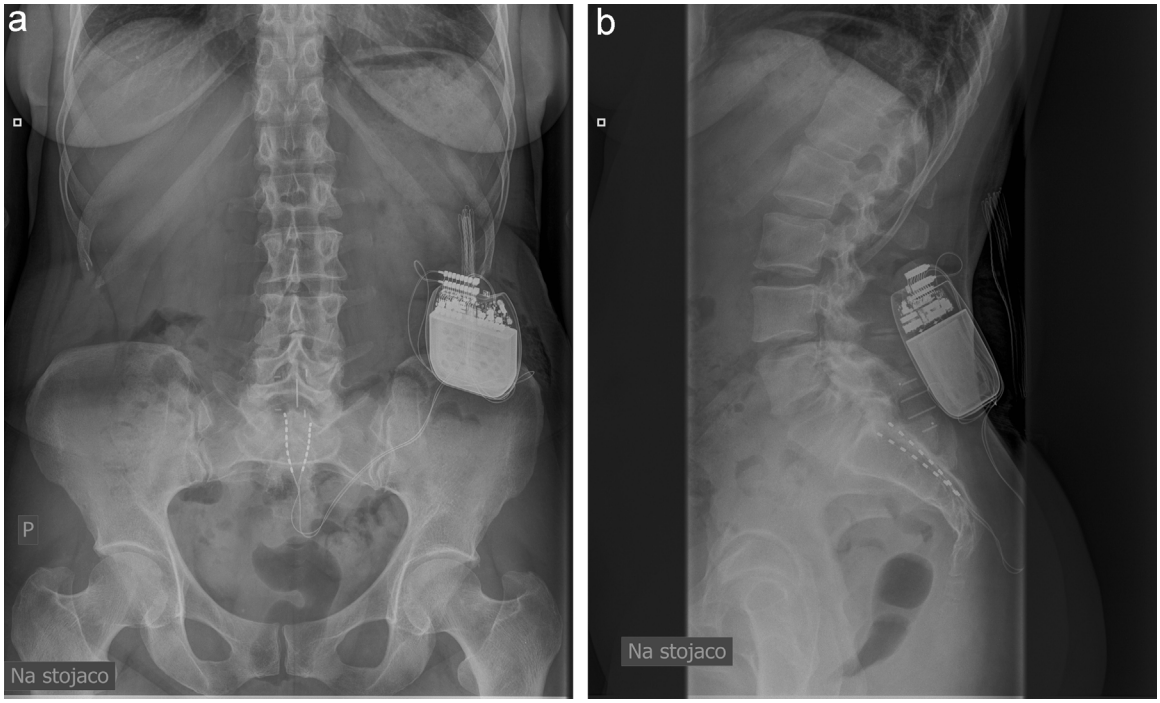

Fig. 2 – (a and b) Two octopolar electrodes. A 55-year-old female with perianal and coccygeal pain exacerbating during stool excretion and menstrual bleeding or sitting radiating to buttock bilaterally for 14 years after traumatic coccygeal injury. She was taking tramadol 600 mg/day, oxycodin 2×/day and gabapentin 600 mg/day. Reduction of pain 50%.

sacral hiatus in seven cases operated in general anesthesia and in two cases in local anesthesia.

4. Operative technique

The surgery can be performed either in general or local anesthesia. A patient is in prone position. The implantation of the electrode(s) through the sacral hiatus and insertion it into the vertebral canal in order to cross the S4, S3, S2 sacral roots is possible in local anesthesia under fluoroscopic control. After the introduction of percutaneous, cylindrical usually octopolar or quadripolar leads into the sacral canal external stimulation can be performed to estimate the coverage and the efficacy of trial stimulation (Figs. 1 and 2a,b). Later on advancement of connecting lead and placement of implantable pulse generator in the subcutaneous pocket in lumbar or buttock area can be conducted preferably in general anesthesia.

5. Results

All nine patients were females suffering on CPP: perineal, anal, perirectal, pudendal or coccygeal region pain. The origin of pain in four cases (44%) lies in the remote effects of lumbar surgery due to discopathy known as failed-back surgery syndrome (FBSS), in other four cases (56%) we have idiopathic CPP or posttraumatic, with sensory or thermal disturbances in the area of pain filling the criteria of diagnosis of CRPS [7] (Table 1). All patients had a statistically significant improvement after the surgery (median VAS = 9 before surgery) (median VAS = 2 after implantation, $p = 0.001$), a good result was also observed after 6 months of observation (median VAS = 3, $p = 0.043$). The long-term follow-up revealed a less satisfactory result and exacerbation of pain in majority of patients (median VAS = 6 after 12 months) (Fig. 3). All patients noticed the improvement of life comfort and felt discomfort after switching off the implanted stimulator. Almost all patients (8/9) reduced analgesic medication. The patient with fecal incontinence reported the reduction of these symptoms. The high incidence of complications was noted: mainly infection – present in three patients and migration of electrodes in two others (Fig. 4).

6. Discussion

In this paper we report a non-lesioning and neuromodulating method of stimulation of sacral roots in order to treat CPP. This is a minimally invasive technique of the percutaneous implantation of electrodes into the sacral canal through the sacral hiatus. Stimulation is reversible and always can be turned off.

In our series we included female patients suffering of CPP of a different origin. The patients' complaints were refractory to conservative management. These patients are difficult to manage due to multiple causes and multiple pathways for pain transmission from the pelvis. The greatest number of cases were patients with perineal, anal and coccygeal pain. Predominant painful dermatomes were S4, S5 in some cases also S3, S2 and S1. The main target of neuromodulation was to

Table 1 – Characteristics of patients.

Nr	Initials	Age	duration of pain in years	VAS before surgery	Sort of pain	electrodes	VAS after SCS	VAS after SRS	VAS after 6 months	VAS after 12 months	Medication	Follow-up
1	BB	43	5	6	Perineal FBSS	2 × 4	5	2	3	–	Reduction of tramadol	After 6 months infection removal
2	HJ	62	6	8	Perineal and coccygeal FBSS	1 × 4	8	2	3	7	Tramadol not reduced	4 years – no medication
3	US	64	17	9	Perineal FBSS	2 × 4	–	4	4	4	Reduction of tramadol	1 year – electrode migration
4	EP	57	6	9	CRPS idiopathic perineal	2 × 4	–	1	–	–	Reduction of gabapentin, withdrawal of tramadol	After 1 m infection removal
5	HP	77	12	9	CRPS idiopathic vulvodynia	1 × 8	–	1	2	6	Carbamazepinum withdrawal and tramadol reduction	18 months migration
6	EM	55	14	9	Perineal and coccygeal CRPS	2 × 8	–	3	3	–	Withdrawal of oxycodon and gabapentin, reduction of tramadol	8 months migration and infection-removal
7	IM	41	5	9	Coccycodynia and left leg	1 × 8	–	1	3	3	Codeine/paracetamol withdrawal	12 months
8	EK	61	5	8	Perineal and perirectal pain CRPS	2 × 8	–	2	3	4	Withdrawal of gabapentin and reduction of oxycodon	12 months
9	MU	48	9	8	Perineal FBSS and fecal incontinence	2 × 8	–	1	1	3	Reduction of tramadol	12 months

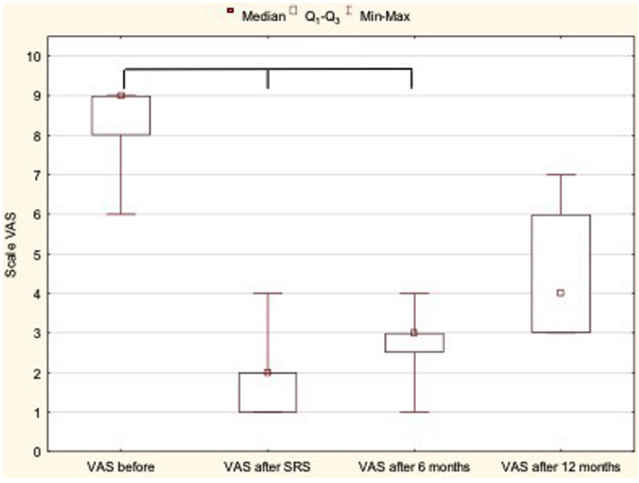

Fig. 3 – Statistically significant improvement after surgery and after 6 months assessed in VAS score.

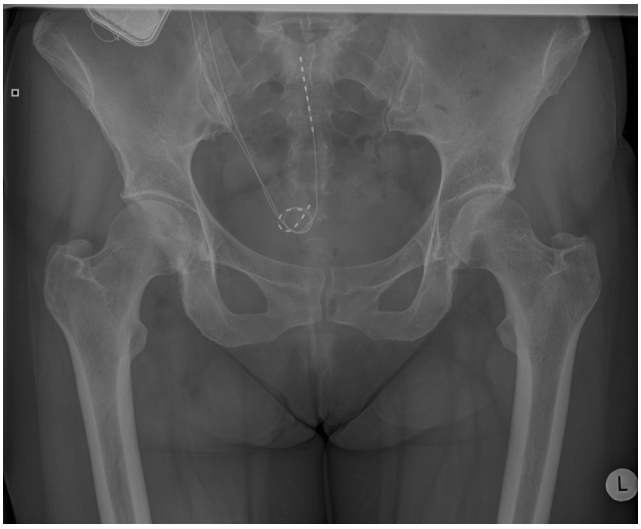

Fig. 4 – One octopolar electrode migrated from the sacral canal. Due to local, purulent infection two electrodes and IPG were removed after 9 months.

cover the greatest possible area of pain and to decrease the intensity of pain at least 50% [17]. At the beginning we tried to use spinal conus stimulation for proper coverage of pain area. Due to insufficient pain relief we had to replace electrodes into the sacral canal to stimulate lower sacral roots in two cases with successful long-term result. The improvement was noticeable. Hunter et al. [3] demonstrated case series of patients with CPP treated successfully with SCS and lead placement at the T6–T7 regions reaching adequate coverage. In our series we do not have patients with CPP treated with T6–T7 SCS but we have two cases with spinal conus stimulation on the level T12-L2. Hunter had one patient with spinal cord stimulation who received good result although he noticed that capture with stimulation of this region is difficult to achieve due to high mobility of the conus under dural sac [3]. In our opinion, nowadays it is easier to stimulate sacral fibers in dorsal columns of spinal cord having availability of, for instance, a five-column electrodes which are dedicated to SCS. Alo and McKay used two sacral nerve root electrodes to stimulate S2–S3 roots bilaterally with excellent result in pelvic pain in interstitial cystitis [18]. Hope presented a case of successful treatment of pelvic pain with SN with electrode implanted at S3 level [1]. All patients after qualification had one stage surgery with permanently implanted IPG (implantable pulse generator) although we usually ask patients whether they prefer trial stimulation. In two cases we performed the implantation in local anesthesia and external trial stimulation was conducted to find the most optimal localization of electrodes leads. One case was complicated with infection 1 month after surgery which ended with removal of pulse generator device and electrodes, although the patient had significant improvement for a month. The patients report excellent relief of their CPP during first months after the implantation ranging from over 50% to 90% diminished intensity of pain. Longer-term results are not so satisfying and after 1 year we should by happy if the patient reaches 50% improvement. The late failure of neuromodulation is the result of development of true tolerance and not always accurate assessment by patients degree of pain relief [17]. Since we did not have trial stimulations we did not excluded any patients from permanent implants and that is why this group better reflects the efficacy of this method in the treatment of CPP. We have to admit that we have high complication rate associated with infection despite the fact that we meticulously follow the rules of antiseptic and aseptic conditions of surgery. Bendersky i Yampolski reviewed complications in SCS and presented recommendations allowing on the reduction of the infection rate. Among them there were antimicrobial prophylaxis before surgery, at least double change of gloves, aseptic conditions, limited use of electrocautery, rinsing of wounds with aqueous povidone–iodine solution, nonstriangulating sutures and avoidance of placing devices or cables under incision line [19]. The last condition in this method is difficult to fulfill. Three cases of electrode migration shows that this technique has its shortcomings associated with difficulty of fixing the electrode near the coccygeal bone under the skin devoid of muscles and fascia which could enable strong fixation. Cases of infection were treated at the beginning conservatively, after failed antibiotic therapy devices had to be removed. Feler et al. [20] presented opinion that retrograde (directed caudally) implantation of electrodes offers superior effectiveness with fewer complications comparing to other methods of sacral neuromodulation. The objective of proper neurostimulation is to achieve correct coverage of the pain area with stimulation paresthesias and in this way substantial and durable pain relief. Perineal, anal, rectal and tailbone area can be covered by SRS. Parameters modification changes sensation of induced paresthesia. It increases efficacy of stimulation and contributes to further improvement of comfort of life and pain relief. Eventual interruption of stimulation helps the patients realize and appreciate the value of this treatment [21]. It seems that SRS should not be considered a panaceum for CPP but can be a part of treatment ladder [22]. Unfortunately the procedure is burdened with the increased risk of complications basing on

our experience and it must be kept in mind when treating patients with CPP with this method.

The main limitation of this paper lies in a small number of observed cases and lack of a control group and no other outcome measures apart from visual analog scale. There are trials, which were conducted in fecal and urinary incontinence or interstitial cystitis but there are no many reports on series of patients with perianal and coccygeal pain of another origin treated with neuromodulation.

7. Conclusions

SRS could be a promising technique in patients with CPP. The short-term therapeutic effects of this intervention are satisfactory. It is non-destructive and minimally invasive method. It can be effective even in the long-term observation but particular care is advised to secure aseptic conditions in the implantation and prevent the infection which leads to eventual removal of the system.

Conflict of interest statement

None declared.

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None declared.

Ethics

The work described in this article has been carried out in accordance with The Code of Ethics of the World Medical Association (Declaration of Helsinki) for experiments involving humans; Uniform Requirements for manuscripts submitted to Biomedical journals.

REFERENCES

[1] Hope ER, Gruber DD. Coccygeal fracture pain cured by sacral neuromodulation: a case report. Neuromodulation 2013;16:614–7.
[2] Patijn J, Janssen M, Hayek S, Mekhail N, Van Zundert J, van Kleef M. Coccygodynia. Pain Pract 2010;10:554–9.
[3] Hunter C, Davé N, Diwan S, Deer S. Neuromodulation of pelvic visceral pain: review of the literature and case series of potential novel targets for treatment. Pain Pract 2013;13:3–17.
[4] Kothari S. Neuromodulatory approaches to chronic pelvic pain and coccycodynia. Act Neurochir Suppl 2007;97(1):365–71.
[5] Malec-Milewska M, Horosz B, Kolęda I, Sękowska A, Kucia H, Kosson D, et al. Case report: Neurolytic block of ganglion of Walther for the management of chronic pelvic pain. Videosurg Miniinv 2014;9:458–62.
[6] Janicki TI. Chronic pelvic pain as a form of complex regional pain syndrome. Clin Obstet Gynecol 2003;46:797–803.
[7] Harden RN, Bruehl S, Stanton-Hicks M, Wilson PR. Proposed new diagnostic criteria for complex regional pain syndrome. Pain Med 2007;8:326–31.
[8] Al Tamimi M, Davids HR, Barolat G, Krutsch J, Ford T. Subcutaneous peripheral nerve stimulation treatment for chronic pelvic pain. Neuromodulation 2008;11:277–81.
[9] Bennett DS, Brookoff D. Sacral nerve root stimulation for interstitial cystitis. In: Simpson BA, editor. Electrical stimulation and the relief of pain. Elsevier; 2003. p. 57–77.
[10] Sherman ND, Amundsen CL. Current and future techniques of neuromodulation for bladder dysfunction. Curr Urol Rep 2007;8:448–554.
[11] Schreiner L, Santos TG, Souza AB, Nygaard CC, Silva Filho IG. Electrical stimulation for urinary incontinence in woman: a systematic review. Int Braz J Urol 2013;39:454–64.
[12] Marcelissen T, Jacobs R, van Kerrebroeck P, de Wachter S. Sacral neuromodulation as a treatment for chronic pelvic pain. J Urol 2011;186(2):387–93.
[13] Siegel S, Noblett K, Mangel J, Griebling TL, Sutherland SE, Bird ET, et al. Results of a prospective, randomized, multicenter study evaluating sacral neuromodulation with InterStim therapy compared to standard medical therapy at 6-months in subjects with mild symptoms of overactive bladder. Neurourol Urodyn 2015;34:224–30. http://dx.doi.org/10.1002/nau.22544
[14] Peters KM, Feber KM, Bennett RC. A prospective, single blind, randomized crossover trial of sacral vs pudendal nerve stimulation for interstitial cystitis. BJU Int 2007;100:835–9.
[15] Srivastava D. Efficacy of sacral neuromodulation in treating chronic pain related to painful bladder syndrome/interstitial cystitis in adults. J Anaesthesiol Clin Pharmacol 2012;28:428–35.
[16] Melzack R, Wall PD. Pain mechanisms: a new theory. Science 1965;150:971–9.
[17] Simpson B. Selection of patients and assessment of outcome. In: Simpson BA, editor. Electrical stimulation and the relief of pain. Elsevier; 2003. p. 237–49.
[18] Alo KM, Mckay E. Selective nerve root stimulation (SNRS) for the treatment of intractable pelvic pain and motor dysfunction: a case report. Neuromodulation 2001;4:19–23.
[19] Bendersky D, Yampolsky C. Is spinal cord stimulation safe? A review of its complications. World Neurosurg 2014;82:1359–68. http://dx.doi.org/10.1016/j.wneu.2013.06.012
[20] Feler CA, Whitworth LA, Fernandez J. Sacral neuromodulation for chronic pain conditions. Anesthesiol Clin N Am 2003;21:785–95.
[21] Sokal P, Harat M, Paczkowski D, Rudaś M, Birski M, Litwinowicz A. Results of neuromodulation for the management of chronic pain. Neurol Neurochir Pol 2011;45:445–51.
[22] Puylaert M. Pelvic pain: mechanistically enigmatic therapeutically challenging. Pain Pract 2012;13:1–2.